**Benefits and harms adopted in health economic assessments evaluating antenatal and newborn screening programmes in the OECD countries: A systematic review of 336 articles and reports**

**Supplementary Material**

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### Supplementary Table 1. Search strategy for Ovid MEDLINE® Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE® Daily and Ovid MEDLINE® 1946 to present

Search Strategy: A full search of the published literature was conducted on 24th April 2020 with a top-up search conducted on 2nd July 2020 to include the search term ‘perinatal’, and a refresh search on 22nd January 2021.

| **#** | **Query** |
| --- | --- |
| 1 | Ultrasonography, Prenatal/ |
| 2 | exp Prenatal diagnosis/ |
| 3 | (ultrasound\* or ultra-sound or ultrasonogra\* or ultra-sonogra\* or sonogra\* or echocardiogra\* or nuchal translucen\* or amniocentesis or chorionic villus sampl\* or cvs or (((noninvasive prenatal or non-invasive prenatal) adj2 (test\* or screen\*)) or nipt)).ti,ab. |
| 4 | ((fetal or foetal or fetus or foetus or prenat\* or pre-nat\* or prepart\* or pre-part\* or antenatal or ante-natal or perinatal or pregnant or pregnancy or trimester?) adj3 (screen\* or test\* or diagnos\* or scan\* or structural assessment\* or structural survey\*)).ti,ab. |
| 5 | screen\*.ti. |
| 6 | exp Abortion, Induced/ |
| 7 | ((induced or therap\*) adj3 abortion?).ab,kw. or abortion?.ti. |
| 8 | 1 or 2 or 3 or 4 or 5 or 6 or 7 |
| 9 | exp Congenital Abnormalities/ |
| 10 | primary dysautonomias/ or dysautonomia, familial/ or Tay-Sachs Disease/ |
| 11 | Muscular Atrophy, Spinal/ |
| 12 | (dysautonomia? or tay sachs).ti,ab,kw. |
| 13 | (congenital\* adj2 (defect? or malformation? or abnormalit\* or anomal\* or aneuploid\*)).ti,ab. |
| 14 | ((fetal or foetal or fetus or foetus) adj2 (defect? or malformation? or abnormalit\* or anomal\* or aneuploid\*)).ti,ab. |
| 15 | ((structural or neural tube?) adj2 (defect? or malformation? or abnormalit\* or anomal\*)).ti,ab. |
| 16 | ((non-chromosom\* or nonchromosom\* or chromosom\*) adj2 (defect? or malformation? or abnormalit\* or anomal\*)).ti,ab. |
| 17 | (((down\* or patau\* or edward\*) adj2 syndrome\*) or trisomy 13 or trisomy 18 or trisomy 21).ti,ab. |
| 18 | spinal muscular atrophy.ti,ab,kw. |
| 19 | 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 |
| 20 | 8 and 19 |
| 21 | exp Congenital Abnormalities/di, dg |
| 22 | Prenatal Care/ or Perinatal Care/ |
| 23 | (fetal or foetal or fetus or foetus or prenat\* or pre-nat\* or prepart\* or pre-part\* or antenatal or ante-natal or perinatal or pregnant or pregnancy or trimester?).ti,ab. |
| 24 | 22 or 23 |
| 25 | 21 and 24 |
| 26 | ((fetal or foetal or fetus or foetus or prenat\* or pre-nat\* or prepart\* or pre-part\* or antenatal or ante-natal or perinatal) adj (screen\* or test\* or diagnos\*)).ti. |
| 27 | 20 or 25 or 26 |
| 28 | Diabetes, Gestational/ or exp Hypertension, Pregnancy-Induced/ |
| 29 | (eclampsia or preeclampsia or pregnancy induced hypertension).ti,ab,kw. |
| 30 | ((gestational or pregnan\* or maternal) adj2 diabet\*).ti,ab,kw. |
| 31 | exp Obstetric Labor, Premature/ or Vasa Previa/ or Placenta Previa/ or Fetal Death/ |
| 32 | ((preterm or premature) adj2 labo?r).ti,ab,kw. |
| 33 | (f?etal death? or stillbirth? or still birth?).ti,ab,kw. |
| 34 | ((placenta or vasa) adj pr?evia).ti,ab,kw. |
| 35 | anemia, hemolytic, congenital/ or exp anemia, sickle cell/ or exp thalassemia/ |
| 36 | (sickle cell or thalass?emia?).ti,ab,kw. |
| 37 | exp Syphilis/ |
| 38 | syphilis.ti,ab,kw. |
| 39 | exp Hepatitis B/ |
| 40 | Hepatitis B virus/ |
| 41 | (hepatitis b or hbv).ti,ab,kw. or hepatitis.ti. |
| 42 | exp HIV/ |
| 43 | exp HIV Infections/ |
| 44 | (hiv or human immunodeficiency virus).ti,ab,kw. |
| 45 | exp Chlamydia Infections/ or exp Chlamydia/ |
| 46 | chlamydia.ti,ab,kw. |
| 47 | exp Cytomegalovirus Infections/ |
| 48 | cytomegalovirus.ti,ab,kw. |
| 49 | exp Streptococcal Infections/ |
| 50 | (group b strep or strep b or (streptococc\* adj infection?)).ti,ab,kw. |
| 51 | exp Parvoviridae Infections/ |
| 52 | parovirus.ti,ab,kw. |
| 53 | Rubella/ or Rubella virus/ or Rubella Syndrome, Congenital/ |
| 54 | rubella.ti,ab,kw. |
| 55 | Toxoplasmosis/ or Toxoplasmosis, Congenital/ |
| 56 | toxoplasmosis.ti,ab,kw. |
| 57 | exp Anemia/ |
| 58 | exp Blood Group Antigens/ |
| 59 | exp Thrombophilia/ |
| 60 | thrombophilia?.ti,ab,kw. |
| 61 | an?emia?.ti,ab,kw. |
| 62 | (blood group? or rhd status or rhesus positive or rhesus negative or rhesus status).ti,ab,kw. |
| 63 | exp Urinary Tract Infections/ |
| 64 | ("urinary tract infection\*" or "urine infection\*" or uti or cystitis or bacteriuria).ti,ab,kw. |
| 65 | Vaginosis, Bacterial/ |
| 66 | vaginosis.ti,ab,kw. |
| 67 | domestic violence/ or spouse abuse/ |
| 68 | ((spous\* or intimate partner or domestic) adj2 (violence or abuse)).ti,ab,kw. |
| 69 | or/28-68 |
| 70 | exp pregnancy/ or pregnant women/ |
| 71 | exp fetus/ |
| 72 | (pregnan$ or f?etal or f?etus or FVS).ti,ab. |
| 73 | preconception care/ or prenatal care/ or perinatal care/ |
| 74 | (pregnan\* or preconception\* or pre-conception\* or antenat\* or ante-nat\* or antepart\* or ante-part\* or prenat\* or pre-nat\* or prepart\* or pre-part\* or perinatal or maternal or mother\*).ti,ab. |
| 75 | 70 or 71 or 72 or 73 or 74 |
| 76 | Mass Screening/ |
| 77 | screen\*.ti,ab. |
| 78 | exp Population Surveillance/ |
| 79 | Self Report/ |
| 80 | (selfreport\* or self-report\* or ((oral or tak\*) adj3 history)).ti,ab. |
| 81 | exp Hematologic Tests/ or Diagnostic Tests, Routine/ or Serologic Tests/ |
| 82 | ((h?ematolog\* or blood or serum or serologic\*) adj3 (test\* or assay\*)).ti,ab. |
| 83 | ((sero\* adj5 (test\* or screen\* or diagnos\*)) or (serotest\* or seroscreen\* or serodiagnos\*)).ti,ab. |
| 84 | exp immunoassays/ |
| 85 | Polymerase Chain Reaction/ |
| 86 | (immuno-assay\* or immunoassay\* or elisa or eia or Fluorescent antibody to membrane antibod\* or fama or trfia).ti,ab. |
| 87 | (enzyme linked immunosorbent assay\* or elisa or enzyme immunoassay\* or eia or recombinant immunoblot assay\* or riba).ti,ab. |
| 88 | (polymerase chain reaction or pcr).ti,ab. |
| 89 | (routine adj5 (test\* or screen\* or diagnos\*)).ti,ab. |
| 90 | (test\* or diagnos\* or assay\*).ti. |
| 91 | 76 or 77 or 78 or 79 or 80 or 81 or 82 or 83 or 84 or 85 or 86 or 87 or 88 or 89 or 90 |
| 92 | 69 and 75 and 91 |
| 93 | Prenatal diagnosis/ or maternal serum screening tests/ |
| 94 | ((pregnan\* or preconception\* or pre-conception\* or antenat\* or ante-nat\* or antepart\* or ante-part\* or prenat\* or pre-nat\* or prepart\* or pre-part\* or perinatal or maternal or mother\*) adj5 (screen\* or diagnos\* or test\*)).ti,ab. |
| 95 | 93 or 94 |
| 96 | 69 and 95 |
| 97 | Diabetes, Gestational/di or exp Hypertension, Pregnancy-Induced/di or exp Obstetric Labor, Premature/di or Vasa Previa/di or Placenta Previa/di or Fetal Death/di or anemia, hemolytic, congenital/di or exp anemia, sickle cell/di or exp thalassemia/di |
| 98 | exp Syphilis/di or exp Hepatitis B/di or exp HIV Infections/di or exp Chlamydia Infections/di or exp Cytomegalovirus Infections/di or exp Streptococcal Infections/di or exp Parvoviridae Infections/di or Rubella/di or Rubella Syndrome, Congenital/di or Toxoplasmosis/di or Toxoplasmosis, Congenital/di |
| 99 | exp Anemia/di or exp Blood Group Antigens/di or exp Thrombophilia/di |
| 100 | exp Urinary Tract Infections/di or Vaginosis, Bacterial/di |
| 101 | 97 or 98 or 99 or 100 |
| 102 | 75 and 101 |
| 103 | 92 or 96 or 102 |
| 104 | Neonatal Screening/ |
| 105 | (heelprick\* or heel prick\*).ti,ab,kw. |
| 106 | ((neonat\* or newborn) adj2 screen\*).ti,ab,kw. |
| 107 | exp Infant, Newborn/ |
| 108 | (newborn? or neonat\* or infant?).ti,ab,kw. |
| 109 | 107 or 108 |
| 110 | Physical Examination/ |
| 111 | (physical adj3 exam\*).ti,ab,kw. |
| 112 | Mass screening/ |
| 113 | screen\*.ti,ab,kw. |
| 114 | Genetic testing/ |
| 115 | early diagnosis/ |
| 116 | diagnostic tests, routine/ |
| 117 | (routine adj5 (test\* or diagnos\*)).ti,ab,kw. |
| 118 | Serologic Tests/ |
| 119 | serologic.ti,ab,kw. |
| 120 | ((sero\* adj5 diagnos\*) or (serotest\* or seroscreen\* or serodiagnos\*)).ti,ab,kw. |
| 121 | Dried Blood Spot Testing/ |
| 122 | (blood spot\* or bloodspot\*).ti,ab,kw. |
| 123 | exp Hearing Tests/ |
| 124 | ((hearing or auditor\* or acoustic\* or otoacoustic\*) adj3 (test\* or diagnos\*)).ti,ab,kw. |
| 125 | (automated auditory brainstem response? or aabr or otoacoustic emission? or aoae).ti,ab,kw. |
| 126 | 110 or 111 or 112 or 113 or 114 or 115 or 116 or 117 or 118 or 119 or 120 or 121 or 122 or 123 or 124 or 125 |
| 127 | anemia, hemolytic, congenital/ or anemia, sickle cell/ or exp thalassemia/ |
| 128 | (sickle cell or thalass?emia?).ti,ab,kw. |
| 129 | eye diseases/ or eye diseases, hereditary/ or cataract/ or vision disorders/ or exp blindness/ |
| 130 | (cataract? or blind\* or ((eye? or vision?) adj2 (disease? or disorder?))).ti,ab,kw. |
| 131 | exp Heart Defects, Congenital/ |
| 132 | ((heart or cardi\* or septal or atrial or ventric\*) adj2 (defect? or anomal\* or malformation?)).ti,ab,kw. |
| 133 | ((coarctat\* adj2 aorta) or (valv\* adj2 stenosis) or "transdisposition of the great arter\*" or patent ductus arteriosus or ebstein\* anomal\* or "tetralogy of fallot" or hypoplastic left heart syndrome or tricuspid atresia or truncus arteriosus or anomalous pulmonary venous connection).ti,ab,kw. |
| 134 | Hip Dislocation/ |
| 135 | (hip? adj2 (dysplasia? or dislocat\*)).ti,ab,kw. |
| 136 | exp testicular diseases/ |
| 137 | (((undescend\* or retract\*) adj2 testic\*) or cryptorchid\*).ti,ab,kw. |
| 138 | exp Hearing Loss/ |
| 139 | ((hearing adj2 (loss or disorder?)) or deaf\*).ti,ab,kw. |
| 140 | Cystic Fibrosis/ |
| 141 | cystic fibrosis.ti,ab,kw. |
| 142 | Congenital Hypothyroidism/ |
| 143 | congenital hypothyroid\*.ti,ab,kw. |
| 144 | Biliary Atresia/ |
| 145 | biliary atresia.ti,ab,kw. |
| 146 | exp Genetic Diseases, Inborn/ or exp "Sex Chromosome Disorders of Sex Development"/ |
| 147 | Muscular Dystrophy, Duchenne/ or exp Muscular Atrophy, Spinal/ |
| 148 | (phenylketonuria? or medium chain acyl coa dehydrogenase deficien\* or medium chain acylcoa dehydrogenase deficien\* or mcadd or maple syrup urine disease? or msud or isovaleric acid?emia? or iso-valeric acid?emia? or glutaric aciduria? or homocystinuria? or amino acid metabolism disorder? or biotinidase deficiency or congenital adrenal hyperplasia or duchenne muscular dystrophy or oxidation disorder? or thrombocytop?enia? or galactos?emia? or kernicterus or dehydrogenase deficiency or lchadd or mucopolysaccharidosis or severe combined immunodeficienc\* or spinal muscular atrophy or tyrosin?emia? or adrenoleukodystrophy or ccald or canavan or klinefelter syndrome or 22q11 deletion syndrome or digeorge syndrome).ti,ab. |
| 149 | 127 or 128 or 129 or 130 or 131 or 132 or 133 or 134 or 135 or 136 or 137 or 138 or 139 or 140 or 141 or 142 or 143 or 144 or 145 or 146 or 147 or 148 |
| 150 | 109 and 126 and 149 |
| 151 | 104 or 105 or 106 or 150 |
| 152 | 27 or 103 or 151 |
| 153 | Economics/ |
| 154 | exp "costs and cost analysis"/ |
| 155 | Economics, Dental/ |
| 156 | exp economics, hospital/ |
| 157 | Economics, Medical/ |
| 158 | Economics, Nursing/ |
| 159 | Economics, Pharmaceutical/ |
| 160 | (economic$ or cost or costs or costly or costing or price or prices or pricing or pharmacoeconomic$).ti,ab. |
| 161 | (expenditure$ not energy).ti,ab. |
| 162 | value for money.ti,ab. |
| 163 | budget$.ti,ab. |
| 164 | "Value of Life"/ |
| 165 | quality-adjusted life years/ |
| 166 | Decision Theory/ |
| 167 | (financ\* or fiscal or funding or fee\* or charge\* or budget\*).ti,ab. |
| 168 | (value adj2 (money or monetary)).ti,ab. |
| 169 | ("Value of life" or "quality adjusted life year\*" or qaly\* or qald\* or qale\* or "disability adjusted life year\*" or daly).ti,ab. |
| 170 | (short form\* or shortform\*).ti,ab. |
| 171 | (sf6\* or sf-6\* or sf8 or sf-8 or sf10 or sf-10 or sf12 or sf-12 or sf16 or sf-16 or sf20 or sf-20 or sf36 or sf-36).ti,ab. |
| 172 | (euroqol or euro qol or "euro quality of life" or euroqual or euro qual or eq5d or eq-5d).ti,ab. |
| 173 | (AQoL\* or "Assessment of Quality of Life").ti,ab. |
| 174 | ("16D Health Related Quality of Life" or 16D HRQoL or "17D Health Related Quality of Life" or 17D HRQoL).ti,ab. |
| 175 | ("Child Health Utility 9 Dimension" or CHU9D or "CHU-9D").ti,ab. |
| 176 | 15 dimensional instrument.ti,ab. |
| 177 | ("quality of wellbeing\*" or "quality of well being\*" or qwb).ti,ab. |
| 178 | (hye or health\* year equivalent\*).ti,ab. |
| 179 | (health utilit\* or disutilit\*).ti,ab. |
| 180 | (hui or hui1 or hui2 or hui3 or hui4 or hui-4 or hui-1 or hui-2 or hui-3).ti,ab. |
| 181 | (health\* adj2 priorities).ti,ab. |
| 182 | (Adolescent Health Utility Measure or AHUM).ti,ab. |
| 183 | (preference\* adj3 (valu\* or measur\* or health or life or estimat\* or elicit\* or disease or score\* or instrument or instruments)).ti,ab. |
| 184 | "preference based measure of HRQoL".ti,ab. |
| 185 | (willingness adj2 pay).ti,ab. |
| 186 | standard gamble.ti,ab. |
| 187 | (time trade off or time tradeoff or tto).ti,ab. |
| 188 | (vas or visual analog\*).ti,ab. |
| 189 | discrete choice\*.ti,ab. |
| 190 | (utility elicitation or direct elicitation).ti,ab. |
| 191 | scoring algorithm.ti,ab. |
| 192 | best worst scaling.ti,ab. |
| 193 | (multi attribute utility or multiattribute utility).ti,ab. |
| 194 | (markov or monte carlo method).ti,ab. |
| 195 | exp Resource Allocation/ |
| 196 | Health Priorities/ |
| 197 | ((multicriteria or multi-criteria) adj2 (decision or analys\* or decision aid\* or decision making)).ti,ab. |
| 198 | (benefit risk asessment or risk benefit assessment).ti,ab. |
| 199 | weighted product.ti,ab. |
| 200 | ((analytic\* hierarchy or analytic\* network) adj process\*).ti,ab. |
| 201 | ("measuring attractiveness by a categorical based evaluation technique" or "goal programming" or "elimination and choice expressing reality" or ELECTRE or "preference ranking organization method of enrichment evaluation" or PROMETHEE or "technique for order preference by similarity to ideal solution" or TOPSIS or "measuring attractiveness by a categorical based evaluation technique" or MACBETH).ti,ab. |
| 202 | "Accountability for reasonableness".ti,ab. |
| 203 | (decision\* adj (tree\* or model\* or analysis)).ti,ab. |
| 204 | (resource\* adj2 (use\* or utilisation or allocat\*)).ti,ab. |
| 205 | (ration or rationing).ti,ab. |
| 206 | exp Comparative Effectiveness Research/ |
| 207 | Comparative Effectiveness Research.ti,ab. |
| 208 | or/153-207 |
| 209 | 152 and 208 |
| 210 | ((energy or oxygen) adj cost).ti,ab. |
| 211 | (metabolic adj cost).ti,ab. |
| 212 | ((energy or oxygen) adj expenditure).ti,ab. |
| 213 | 210 or 211 or 212 |
| 214 | 209 not 213 |
| 215 | (comment or letter or editorial or historical article).pt. |
| 216 | 214 not 215 |
| 217 | exp animals/ not humans/ |
| 218 | 216 not 217 |
| 219 | limit 218 to yr="2000 -Current" |
| 220 | (2020\* or 2021\*).ed,ez,yr. |
| 221 | 219 and 220 |

### Supplementary Table 2: List of studies excluded at full-text screening stage, with brief reasons

|  |  |
| --- | --- |
| **Reason of exclusion** | **Source** |
| Other study design (e.g. reviews) | 1–201 |
| Non-economic evaluations | 202–256 |
| Conference abstract that has journal article published | 70,257–310 |
| Non-screening (i.e. not related to antenatal or newborn screening) | 311–333 |
| No details of economic evaluation presented in Methods/Results | 334–343 |
| Duplicate | 344–350 |
| Health preference studies | 351,352 |

### Supplementary Table 3: Characteristics of screening programmes and population in the articles and reports

|  | **Articles and reports assessing antenatal screening (%)** | **Articles and reports assessing newborn screening (%)** | **Total articles and reports (%)** |
| --- | --- | --- | --- |
| Type of screening | 242 (100) | 95 (100) | 337 (100) |
| Setting of screening\* |  |  |  |
| Home | 0 (0) | 3 (3.2) | 3 (0.9) |
| Primary care | 6 (2.5) | 3 (3.2) | 9 (2.7) |
| Secondary care | 58 (24.0) | 22 (23.2) | 80 (23.7) |
| Primary and secondary care | 5 (2.1) | 4 (4.2) | 9 (2.7) |
| Not stated | 173 (71.5) | 63 (66.3) | 236 (70.0) |
| Population\* |  |  |  |
| Healthy pregnancy | 196 (79.0) | 1 (1.0) | 197 (57.1) |
| Pregnant women and their partner/relative | 8 (3.2) | 0 (0) | 8 (2.3) |
| Pregnancy at risk | 37 (14.9) | 1 (1.0) | 38 (11.0) |
| Healthy infant | 7 (2.8) | 84 (86.6) | 91 (26.4) |
| Infant at risk | 0 (0) | 11 (11.3) | 11 (3.2) |
| Gestation stage of pregnant women |  |  |  |
| First trimester | 25 (9.7) | 0 (0) | 25 (7.2) |
| First or second trimester | 3 (1.2) | 0 (0) | 3 (0.9) |
| First and second trimesters | 6 (2.3) | 0 (0) | 6 (1.7) |
| First and third trimesters | 1 (0.4) | 0 (0) | 1 (0.3) |
| Second trimester | 25 (9.7) | 0 (0) | 25 (7.2) |
| Second and third trimesters | 10 (3.9) | 0 (0) | 10 (2.9) |
| Third trimester | 19 (7.4) | 0 (0) | 19 (5.5) |
| Not stated | 168 (65.4) | 2 (2.2) | 170 (49.0) |
| Not applicable | 0 (0) | 88 (97.8) | 88 (25.4) |
| Stage of disease pathway |  |  |  |
| Person at risk but no pathological changes present | 24 (9.9) | 6 (6.3) | 30 (8.9) |
| Symptomless stage with pathologically definable change present | 217 (89.7) | 86 (90.5) | 303 (89.9) |
| Signs and/or symptoms exist but condition undiagnosed | 1 (0.4) | 2 (2.1) | 3 (0.9) |
| Clinical phase | 0 (0) | 1 (1.1) | 1 (0.3) |
| Phase(s) of screening programme |  |  |  |
| Screening and diagnostic | 60 (24.8) | 16 (16.8) | 76 (22.6) |
| Screening and intervention | 59 (24.4) | 10 (10.5) | 69 (20.5) |
| Screening, diagnostic and intervention | 103 (42.6) | 59 (62.1) | 162 (48.1) |
| Not clear | 20 (8.3) | 10 (10.5) | 30 (8.9) |

\* Will not total up to 100% as some articles and reports presented more than one category.

### Supplementary Table 4: Medical conditions investigated

|  |  |  |
| --- | --- | --- |
| **Conditions** | **Articles and reports assessing antenatal screening (%)** | **Articles and reports assessing newborn screening (%)** |
| Developmental | 0 (0) | 6 (6.3) |
| Endocrinology | 24 (9.9) | 4 (4.2) |
| Genetic | 77 (31.8) | 12 (12.6) |
| Gestational cardiorenal | 5 (2.1) | 0 (0) |
| Haematology | 18 (7.4) | 12 (12.6) |
| Infection | 76 (31.4) | 3 (3.2) |
| Intrauterine fetal demise/ sudden infant death syndrome | 1 (0.4) | 1 (1.1) |
| Maternal mental health | 1 (0.4) | 6 (6.3) |
| Metabolic | 0 (0) | 32 (33.7) |
| Neurodevelopment | 1 (0.4) | 3 (3.2) |
| Neurological | 0 (0) | 1 (1.1) |
| Nutrition | 4 (1.7) | 0 (0) |
| Structural | 36 (14.9) | 25 (26.3) |
| Urology | 1 (0.4) | 0 (0) |
| All conditions\* | 242 | 95 |

\*The sum of the individual conditions is not equivalent to the total value across all conditions because there are five articles and reports that investigated more than one condition.

### Supplementary Table 5: Summary of reporting quality of articles and reports (excluding conference abstracts) assessed using CHEERS checklist

| **CHEERS item no.** | **CHEERS item** | **Articles and reports assessing antenatal screening (%)** | | | **Articles and reports assessing newborn screening (%)** | | |
| --- | --- | --- | --- | --- | --- | --- | --- |
| **Satisfied** | **Not satisfied** | **Not applicable** | **Satisfied** | **Not satisfied** | **Not applicable** |
|  | ***Title and abstract*** |  |  |  |  |  |  |
| 1 | Title | 155 (85.6) | 26 (14.4) | 0 (0) | 70 (86.4) | 11 (13.6) | 0 (0) |
| 2 | Abstract | 21 (11.6) | 160 (88.4) | 0 (0) | 13 (15.7) | 69 (83.1) | 1 (1.2) |
|  | ***Introduction*** |  |  |  |  |  |  |
| 3 | Background and objectives | 181 (100) | 0 (0) | 0 (0) | 83 (100) | 0 (0) | 0 (0) |
|  | ***Methods*** |  |  |  |  |  |  |
| 4 | Target population and subgroups | 181 (100) | 0 (0) | 0 (0) | 83 (100) | 0 (0) | 0 (0) |
| 5 | Setting and location | 65 (35.9) | 116 (64.1) | 0 (0) | 30 (36.1) | 53 (63.9) | 0 (0) |
| 6 | Study perspective | 134 (74) | 47 (26) | 0 (0) | 67 (80.7) | 15 (18.1) | 1 (1.2) |
| 7 | Comparators | 159 (87.8) | 22 (12.2) | 0 (0) | 75 (90.4) | 8 (9.6) | 0 (0) |
| 8 | Time horizon | 28 (15.5) | 153 (84.5) | 0 (0) | 15 (18.1) | 67 (80.7) | 1 (1.2) |
| 9 | Discount rate | 51 (28.2) | 130 (71.8) | 0 (0) | 29 (34.9) | 53 (63.9) | 1 (1.2) |
| 10 | Choice of health outcomes | 181 (100) | 0 (0) | 0 (0) | 82 (98.8) | 0 (0) | 1 (1.2) |
| 11 | Measurement of effectiveness | 178 (98.3) | 3 (1.7) | 0 (0) | 82 (98.8) | 1 (1.2) | 0 (0) |
| 12 | Measurement and valuation of preference-based outcomes | 48 (26.5) | 34 (18.8) | 99 (54.7) | 19 (22.9) | 13 (15.7) | 51 (61.4) |
| 13 | Estimate resources and cost | 168 (92.8) | 13 (7.2) | 0 (0) | 79 (95.2) | 3 (3.6) | 1 (1.2) |
| 14 | Currency, price date, and conversion | 139 (76.8) | 42 (23.2) | 0 (0) | 64 (77.1) | 18 (21.7) | 1 (1.2) |
| 15 | Choice of model | 28 (15.5) | 153 (84.5) | 0 (0) | 28 (33.7) | 55 (66.3) | 0 (0) |
| 16 | Assumptions | 131 (72.4) | 20 (11) | 30 (16.6) | 60 (72.3) | 6 (7.2) | 17 (20.5) |
| 17 | Analytic method | 116 (64.1) | 65 (35.9) | 0 (0) | 63 (75.9) | 20 (24.1) | 0 (0) |
|  | ***Results*** |  |  |  |  |  |  |
| 18 | Study parameters | 155 (85.6) | 26 (14.4) | 0 (0) | 73 (88) | 9 (10.8) | 1 (1.2) |
| 19 | Incremental costs and outcomes | 168 (92.8) | 13 (7.2) | 0 (0) | 75 (90.4) | 7 (8.4) | 1 (1.2) |
| 20 | Characterizing uncertainty | 161 (89) | 20 (11) | 0 (0) | 69 (83.1) | 13 (15.7) | 1 (1.2) |
| 21 | Characterizing heterogeneity | 24 (13.3) | 3 (1.7) | 154 (85.1) | 4 (4.8) | 0 (0) | 79 (95.2) |
|  | ***Discussion*** |  |  |  |  |  |  |
| 22 | Study funding, limitation, generalisability, and current knowledge | 58 (32) | 123 (68) | 0 (0) | 23 (27.7) | 59 (71.1) | 1 (1.2) |
|  | ***Other*** |  |  |  |  |  |  |
| 23 | Source of funding | 100 (55.2) | 81 (44.8) | 0 (0) | 55 (66.3) | 28 (33.7) | 0 (0) |
| 24 | Conflict of interest | 107 (59.1) | 74 (40.9) | 0 (0) | 51 (61.4) | 32 (38.6) | 0 (0) |

### Supplementary Table 6: Thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes

| **Theme no.** | **Theme** | **Subtheme level 1** | **Subtheme level 2** | **Subtheme level 3** | **Subtheme level 4** | **Antenatal screening** | **Newborn screening** |
| --- | --- | --- | --- | --- | --- | --- | --- |
| 1 | Diagnosis of screened for condition | additional screening of partners |  |  |  | 353 | - |
| 1 | Diagnosis of screened for condition | additional testing to reach diagnosis in the absence of screening (links to diagnostic odyssey) |  |  |  | 354–356 | 357 |
| 1 | Diagnosis of screened for condition | born with condition | reduction in children born with condition through effective treatment of the screened for condition |  |  | 358–380 | 361,381 |
| 1 | Diagnosis of screened for condition | born with condition | reduction in children born with condition through termination of pregnancy |  |  | 382–408 | - |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening |  |  |  | 356,382–384,386,388,391,397,398,408–430 | 357,431–445 |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening rather than later symptomatically |  |  |  | 390,404,406,446–448 | 276,449–455 |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening that would have become symptomatic |  |  |  | - | 456 |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening | maternal |  |  | 385 | - |
| 1 | Diagnosis of screened for condition | cases missed at screening |  |  |  | 367,402,411,416,417,428,429,457,458 | 459,460 |
| 1 | Diagnosis of screened for condition | cases missed at screening | legal cost of reimbursing false negatives |  |  | - | 461 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition |  |  |  | 270,382–384,404,409,462–465 | 357,433,466–470 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition [invasive] |  |  |  | 355,402 | - |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | early vs late |  |  | - | 435 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | unnecessary due to false positive |  |  | 353,356,358,360,362–365,369–371,374–376,378–380,385,388,390,391,393,397,398,402,403,406,410,411,414–416,418,422,423,425,446–448,471–494 | 276,278,283,381,431,432,434,436–445,449,451–454,456,459,461,469,470,495–522 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | unnecessary due to false positive [invasive] |  |  | 355,392,399,400,405,412,413,415,417,419,421,424,426,428,429,457,523–526 | 527 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | unnecessary due to false positive | time spent attending |  | - | 431 |
| 1 | Diagnosis of screened for condition | screened for condition related complications | morbidity |  |  | - | 438 |
| 2 | Life years and health status adjustments | infant life years post birth | DALYs |  |  | 378,482 | 469,506 |
| 2 | Life years and health status adjustments | infant life years post birth | morbidity |  |  | 374,474,528,529 | 444 |
| 2 | Life years and health status adjustments | infant life years post birth | mortality |  |  | 362,373,374,380,387,430,479,494,528,530,531 | 437,444,519 |
| 2 | Life years and health status adjustments | infant life years post birth | QALYs |  |  | 362,374,376,380,387,464,472,473,479,480,483,488,532–548 | 361,443,444,459,469,496,497,500–503,506,510,511,515,521,549–552 |
| 2 | Life years and health status adjustments | infant life years post birth | replacement of affected aborted fetus with subsequent unaffected pregnancy and life |  |  | 407 | - |
| 2 | Life years and health status adjustments | infant life years post birth | years |  |  | 360,364,366,375,385,403,474,486,487,489,530,535,553,554 | 283,357,361,381,435,444,449,461,468,470,497,498,500,504,505,509,512–517,520,522,552,555,556 |
| 2 | Life years and health status adjustments | infant life years post birth | years | monetary value |  | - | 557 |
| 2 | Life years and health status adjustments | maternal life years | mortality |  |  | 369,374 | 558 |
| 2 | Life years and health status adjustments | maternal life years | QALYs |  |  | 270,376,387,394–396,412,418,420,422,430,458,463–465,471,472,474,476,479–481,483–485,487,490,492–494,523,525,529,531,533,534,537,539,541,545–547,559–571 | 278,466,469,508,515,518,519,527,557 |
| 2 | Life years and health status adjustments | maternal life years | QALYs | decrement due to caring for child with screened for condition |  | 572 | - |
| 2 | Life years and health status adjustments | maternal life years | years |  |  | 360,385,403,474,486,487,489,553,554 | 283,361,381,444,468,470,498,500,504,509,512–515,520,522,552 |
| 2 | Life years and health status adjustments | parental QALYs |  |  |  | - | 449,461,495,507 |
| 2 | Life years and health status adjustments | psychological | anxiety from genetic variants of unclear penetrance |  |  | - | 466 |
| 2 | Life years and health status adjustments | psychological | disutility due to knowledge of disease |  |  | 484 | - |
| 2 | Life years and health status adjustments | psychological | disutility due to knowledge of disease in those with positive screening results (stress and anxiety) |  |  | 422 | - |
| 2 | Life years and health status adjustments | psychological | early diagnosis induced anxiety |  |  | - | 466 |
| 2 | Life years and health status adjustments | psychological | false positive anxiety |  |  | 424 | 439,497 |
| 2 | Life years and health status adjustments | psychological | false positive anxiety | parental QALYs |  | - | 461 |
| 2 | Life years and health status adjustments | psychological | genetic variants of unclear penetrance | unclear harms including behavioural changes | QALYs | - | 466 |
| 2 | Life years and health status adjustments | screened for condition associated mortality / treatment associated mortality/other causes mortality |  |  |  | - | 556 |
| 3 | Treatment | additional healthcare post-diagnosis |  |  |  | 359,528 | 455,466,496 |
| 3 | Treatment | comparison of earlier treatment after screen detection and later after symptomatic detection | increase in treatment required for false negative |  |  | 533,572 | 440,468 |
| 3 | Treatment | comparison of earlier treatment after screen detection and later after symptomatic detection | increase in treatment required for false negative | prior to diagnosis |  | - | 510 |
| 3 | Treatment | comparison of earlier treatment after screen detection and later after symptomatic detection | reduction in treatment required |  |  | 354,548,573 | - |
| 3 | Treatment | comparison of earlier treatment after screen detection and later after symptomatic detection | reduction in treatment required | costs |  | 369,370,422,476,488,535,548,560,564,574–576 | 435,444,461,468,498,499,504,508,515,516,520,522 |
| 3 | Treatment | comparison of earlier treatment after screen detection | adverse complications of screened for condition averted |  |  | 353,354,368–370,410,414,465,477,479,482,487,491,533,545–547,554,560,566,568,576–578 | 6,361,456,513,519,579 |
| 3 | Treatment | hospital stay |  |  |  | 478,540,580 | 503,517 |
| 3 | Treatment | missed due to false negative |  |  |  | 371 | - |
| 3 | Treatment | prevention of screened for condition (infectious) | increase in future earning potential |  |  | 580 | - |
| 3 | Treatment | prevention of screened for condition (infectious) | unnecessary prophylaxis in false positives |  |  | 368,371,372,374,376,379,473,488,532,533,543,570,573,576,580 | - |
| 3 | Treatment | prevention of screened for condition (infectious) | unnecessary prophylaxis in false positives | allergic reaction |  | 570 | - |
| 3 | Treatment | psychological | counselling about genetic diagnosis |  |  | 382,397 | - |
| 3 | Treatment | psychological | counselling about screening or confirmatory test | false positive |  | 366,376,405,406,424,543 | - |
| 3 | Treatment | psychological | counselling about screening or confirmatory test |  |  | 353,382,397 | - |
| 3 | Treatment | screened for condition related treatment/management |  |  |  | 270,353,378,382,409,422,458,464,465,475,546,581 | 467–470,556 |
| 3 | Treatment | screened for condition related treatment/management | unnecessary in false positives with no confirmatory test |  |  | 359,360,362–364,367,371,377,380,385,403,422,471,476,479–481,484,487,490,528,529,538–540,542,545,547,548,553,554,561,562,564,567,572,578 | 276,455,461,498,501,502,505,513,517,521,550,551,557,582 |
| 3 | Treatment | treatment related harm | adverse reaction to treatment |  |  | 372,374,480,485,489,491,532,560,570,572 | 283,505,509,513,517,520,583 |
| 3 | Treatment | treatment related harm | antibiotic resistance |  |  | 359 | - |
| 3 | Treatment | treatment related harm | disutility of treatment |  |  | 487 | - |
| 3 | Treatment | treatment related harm | treatment related anxiety |  |  | - | 515 |
| 3 | Treatment | unnecessary due to false positive |  |  |  | 375,474,544,546 | - |
| 4 | Long-term cost associated with screened for condition | cost-savings from averted births of fetuses with anomalies |  |  |  | 354 | - |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost |  |  |  | 353,374,480 | 497 |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost | healthcare and productivity gains |  |  | 407 | - |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost | healthcare and social services |  |  | - | 518 |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost | healthcare, education and social services |  |  | - | 466,502 |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost | social services |  |  | 380 | 515 |
| 4 | Long-term cost associated with screened for condition | direct healthcare and non-healthcare cost | treatment and caregiving |  |  | 403 | 519 |
| 4 | Long-term cost associated with screened for condition | direct healthcare cost |  |  |  | 270,360,363,364,366,369,373,375,376,382,393,399,401,405,407,457,462,464,465,474,477,479,482–484,486–488,493,530–532,534–536,538,543,544,548,553,559,562,567,568,571,572,575,576,580,581 | 381,443,444,449,450,452,459,461,469,470,496,500,501,503,504,510,511,513,514,522,552,556,584,585 |
| 4 | Long-term cost associated with screened for condition | direct non-healthcare cost |  |  |  | 385 | - |
| 4 | Long-term cost associated with screened for condition | direct non-healthcare cost | caregiving |  |  | 384,473,524,586 | 582 |
| 4 | Long-term cost associated with screened for condition | direct non-healthcare cost | child protective services investigation and foster care placements if mothers successfully completed substance abuse treatment |  |  | 530 | - |
| 4 | Long-term cost associated with screened for condition | direct non-healthcare cost | education and social services |  |  | - | 435 |
| 4 | Long-term cost associated with screened for condition | direct non-healthcare cost | social care |  |  | - | 504 |
| 4 | Long-term cost associated with screened for condition | productivity gains |  |  |  | 370,562 | 550 |
| 4 | Long-term cost associated with screened for condition | societal cost |  |  |  | 356,412,425,523,525 | 276,278,495,550 |
| 5 | Overdiagnosis | QALY decrement |  |  |  | 480 | 515 |
| 5 | Overdiagnosis | unnecessary test/treatment |  |  |  | 477,480 | 461,502,515 |
| 6 | Pregnancy loss | spontaneous |  |  |  | 356,359,383,384,387,402,406,427,478,492,572 | - |
| 6 | Pregnancy loss | termination |  |  |  | 355,387,419,430,571 | - |
| 6 | Pregnancy loss | termination | date/trimester |  |  | 397,412,523 | - |
| 6 | Pregnancy loss | termination | of unaffected fetus due to false positive test result |  |  | 381,386–388,390,391,400,405,415,416,418,420,424,427,579 | - |
| 6 | Pregnancy loss | termination | of unaffected fetus due to false positive test result | psychological consequences |  | 419 | - |
| 6 | Pregnancy loss | termination | prevent downstream adverse maternal outcomes |  |  | 526 | - |
| 6 | Pregnancy loss | termination | prevent later miscarriage |  |  | 359,397 | - |
| 6 | Pregnancy loss | termination | psychological consequences |  |  | 419,526,572 | - |
| 6 | Pregnancy loss | treatment/test related |  |  |  | 278,355,356,382–384,388–392,394,395,397,398,400,402,404–406,408,409,411–413,415,416,418–421,424–429,447,448,457,472,492,525,526,548,571,572,579,584 | - |
| 6 | Pregnancy loss | treatment/test related | unaffected |  |  | 355,382 | - |
| 7 | Spillover effects | benefits to parents from child’s diagnosis with genetic condition, through knowledge of their own genetic status |  |  |  | - | 557 |

### Supplementary Table 7: Summary of thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes by conditions

| **Screening type** | **Core theme** | **Developmental** | **Endocrinology** | **Genetic** | **Gestational cardiorenal** | **Haematology** | **Infection** | **Intrauterine fetal demise** | **Maternal mental health** | **Metabolic** | **Neurodevelopment** | **Neurological** | **Nutrition** | **Structural** | **Urology** |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| Antenatal | Total no. of studies | 0 (0%) | 24 (100%) | 77 (100%) | 5 (100%) | 18 (100%) | 76 (100%) | 1 (100%) | 1 (100%) | 0 (0%) | 1 (100%) | 0 (0%) | 4 (100%) | 36 (100%) | 1 (100%) |
| No themes\* | 0 (0%) | 9 (38%) | 24 (31%) | 1 (20%) | 8 (44%) | 22 (29%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 1 (25%) | 16 (44%) | 0 (0%) |
| Theme 1 | 0 (0%) | 8 (33%) | 49 (64%) | 3 (60%) | 7 (39%) | 32 (42%) | 0 (0%) | 1 (100%) | 0 (0%) | 0 (0%) | 0 (0%) | 2 (50%) | 14 (39%) | 1 (100%) |
| Theme 2 | 0 (0%) | 13 (54%) | 14 (18%) | 2 (40%) | 4 (22%) | 38 (50%) | 1 (100%) | 1 (100%) | 0 (0%) | 1 (100%) | 0 (0%) | 2 (50%) | 14 (39%) | 0 (0%) |
| Theme 3 | 0 (0%) | 11 (46%) | 8 (10%) | 4 (80%) | 7 (39%) | 47 (62%) | 1 (100%) | 1 (100%) | 0 (0%) | 0 (0%) | 0 (0%) | 2 (50%) | 7 (19%) | 1 (100%) |
| Theme 4 | 0 (0%) | 4 (17%) | 15 (19%) | 1 (20%) | 3 (17%) | 36 (47%) | 0 (0%) | 0 (0%) | 0 (0%) | 1 (100%) | 0 (0%) | 0 (0%) | 8 (22%) | 1 (100%) |
| Theme 5 | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 2 (3%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) |
| Theme 6 | 0 (0%) | 0 (0%) | 43 (56%) | 0 (0%) | 2 (11%) | 2 (3%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 1 (25%) | 6 (17%) | 0 (0%) |
| Theme 7 | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) |
| Newborn | Total no. of studies | 6 (100%) | 4 (100%) | 12 (100%) | 0 (0%) | 12 (100%) | 3 (100%) | 1 (100%) | 6 (0%) | 32 (100%) | 3 (100%) | 1 (100%) | 0 (0%) | 25 (100%) | 0 (0%) |
| No themes\* | 3 (50%) | 1 (25%) | 1 (8%) | 0 (0%) | 3 (25%) | 0 (0%) | 0 (0%) | 3 (0%) | 9 (28%) | 1 (33%) | 0 (0%) | 0 (0%) | 3 (12%) | 0 (0%) |
| Theme 1 | 2 (33%) | 2 (50%) | 8 (67%) | 0 (0%) | 8 (67%) | 1 (33%) | 1 (100%) | 2 (0%) | 22 (69%) | 1 (33%) | 0 (0%) | 0 (0%) | 19 (76%) | 0 (0%) |
| Theme 2 | 3 (50%) | 1 (25%) | 8 (67%) | 0 (0%) | 9 (75%) | 0 (0%) | 1 (100%) | 3 (0%) | 19 (59%) | 2 (67%) | 1 (100%) | 0 (0%) | 8 (32%) | 0 (0%) |
| Theme 3 | 1 (17%) | 1 (25%) | 3 (25%) | 0 (0%) | 7 (58%) | 1 (33%) | 1 (100%) | 1 (0%) | 14 (44%) | 1 (33%) | 1 (100%) | 0 (0%) | 11 (44%) | 0 (0%) |
| Theme 4 | 1 (17%) | 2 (50%) | 5 (42%) | 0 (0%) | 4 (33%) | 1 (33%) | 1 (100%) | 1 (0%) | 17 (53%) | 1 (33%) | 0 (0%) | 0 (0%) | 7 (28%) | 0 (0%) |
| Theme 5 | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 3 (9%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) |
| Theme 7 | 0 (0%) | 0 (0%) | 1 (8%) | 0 (0%) | 0 (0%) | 2 (67%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) | 0 (0%) |

Theme 1: Diagnosis of screened for condition, Theme 2: Life years and health status adjustments, Theme 3: Treatment, Theme 4: Long-term cost associated with screened for condition, Theme 5: Overdiagnosis, Theme 6: Pregnancy loss, Theme 7: Spillover effects.  
\*Limited information about benefits and harms could be extracted to inform our bespoke form as the majority in this category were conference abstracts.

### Supplementary Table 8: Thematic framework of benefits and harms adopted by health economic assessments evaluating antenatal and newborn screening programmes in the refresh search from 1st January 2021 to 22nd November 2021

| **Theme no.** | **Theme** | **Subtheme level 1** | **Subtheme level 2** | **Subtheme level 3** | **Antenatal screening** | **Newborn screening** |
| --- | --- | --- | --- | --- | --- | --- |
| 1 | Diagnosis of screened for condition | born with condition | reduction in children born with condition through termination of pregnancy |  | 587 |  |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening |  |  | 588–591 |  |
| 1 | Diagnosis of screened for condition | cases diagnosed at screening that would have become symptomatic |  |  |  | 592 |
| 1 | Diagnosis of screened for condition | cases missed at screening |  |  | 588 |  |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | unnecessary due to false positive |  | 591,593,594 | 592 |
| 1 | Diagnosis of screened for condition | confirmatory test and additional tests to reach diagnosis of screened for condition | unnecessary due to false positive [invasive] |  | 588,595 |  |
| 1 | Diagnosis of screened for condition | screened for condition related complications | morbidity |  | 596 |  |
| 2 | Life years and health status adjustments | infant life years post birth | DALYs |  | 596 |  |
| 2 | Life years and health status adjustments | infant life years post birth | morbidity |  | 590,595–597 |  |
| 2 | Life years and health status adjustments | infant life years post birth | mortality |  | 589,590,597,598 |  |
| 2 | Life years and health status adjustments | infant life years post birth | QALYs |  | 593,598 | 592,599 |
| 2 | Life years and health status adjustments | maternal life years | mortality |  | 594 |  |
| 2 | Life years and health status adjustments | maternal life years | QALYs |  | 587,591,594,598 |  |
| 3 | Treatment | comparison of earlier treatment after screen detection and later after symptomatic detection | reduction in treatment required | costs | 598 |  |
| 3 | Treatment | comparison of earlier treatment after screen detection | adverse complications of screened for condition averted |  | 594,597,598 |  |
| 3 | Treatment | hospital stay |  |  | 590 |  |
| 3 | Treatment | treatment related harm | adverse reaction to treatment |  | 597 |  |
| 3 | Treatment | unnecessary due to false positive |  |  | 589,590,594–598 |  |
| 6 | Pregnancy loss | termination of pregnancy |  |  | 588,595 |  |
| 6 | Pregnancy loss | treatment/test related |  |  | 587,595 |  |
| 6 | Pregnancy loss | treatment/test related | unaffected |  | 588 |  |
| 7 | Spillover effects |  |  |  | 593 |  |

*Note: Five studies from refresh search were not presented here as they did not include benefits/harms of antenatal/newborn screening*600–604*.*

## Supplementary Figures

### Supplementary Figure 1: Number of articles and reports published from 2000 to January 2021

Chart, line chart

Description automatically generated

Note: Dotted lines were used to indicate that only January 2021 was included in this chart.

## References of Supplementary Material

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