**Appendix 3**

***Search strategy***

A search was performed on January 16/17 2018 in these databases: Cochrane, Medline, Embase, PsycInfo, Cinahl, Web of Science/Knowledge. Databases and search strategy are given below.

Medline

1 Craniosynostoses/ (4784)

2 craniosynost\*.ti,ab. (4255)

3 craniosynost\*.kf. (606)

4 (synost\* and skull\*).ti,ab. (472)

5 Craniofacial Abnormalities/ (5500)

6 Skull/ab [Abnormalities] (4484)

7 or/1-6 (15229)

8 (dutch or german or english).la. (26393872)

9 (animals not humans).sh. (4781706)

10 7 not 9 (14000)

11 8 and 10 (12459)

12 11 (12459)

13 limit 12 to yr="2009 -Current" (4517)

14 guideline/ or practice guideline/ (32919)

15 guideline?.ti,ab. (301908)

16 guideline?.ti. (70152)

17 guideline?.kf. (9513)

18 14 or 16 or 17 (94185)

19 13 and 18 (10)

20 11 (12459)

21 limit 20 to yr="2000 -Current" (7785)

22 "cradock$".fc\_auts. and "language learning".fc\_titl. and "2016".fc\_pubyr. and "1".fc\_pg. (0)

23 "wallace$".fc\_auts. and "single-suture craniosynostosis".fc\_titl. and "2016".fc\_pubyr. (4)

24 from 23 keep 3 (1)

25 "3".fc\_issue. and "39".fc\_vol. and "knight$".fc\_auts. and "neurodevelopmental outcomes".fc\_titl. (1)

26 from 25 keep 1 (1)

27 language/ or exp speech/ (66744)

28 exp Learning/ (375119)

29 Developmental Disabilities/ (19683)

30 (language or speech).tw. (186150)

31 (language or speech).kf. (14920)

32 (development\* adj3 disabilit\*).tw. (6545)

33 (development\* adj3 disabilit\*).kf. (628)

34 or/27-33 (593506)

35 21 and 34 (568)

36 probability/ or proportional hazards models/ or risk/ or exp risk assessment/ or risk factors/ (1163082)

37 risk.tw,kf. (1903090)

38 36 or 37 (2378732)

39 35 and 38 (64)

40 (language or speech).ti. (51370)

41 (development\* adj3 disabilit\*).ti. (2413)

42 \*language/ or exp \*speech/ (40274)

43 exp \*Learning/ (230251)

44 \*Developmental Disabilities/ (13448)

45 31 or 33 or 40 or 41 or 42 or 43 or 44 (318227)

46 35 and 45 (214)

47 38 and 46 (27)

48 exp Language Disorders/ (49111)

49 exp Language Disorders/et [Etiology] (12960)

50 45 or 49 (327260)

51 35 and 45 (214) 180

52 38 and 51 (27)

53 "filter observational studies Medline".ti. (0)

54 epidemiologic studies/ (8321)

55 exp case-control studies/ (1002942)

56 exp cohort studies/ (1913426)

57 cross-sectional studies/ (285841)

58 (case adj3 control).af. (319523)

59 (cohort adj5 (study or studies or analy$)).af. (382069)

60 (follow-up adj5 (study or studies)).af. (698063)

61 (longitudinal or retrospective or prospective or (cross adj5 sectional)).af. (2137614)

62 (observational adj5 (study or studies)).af. (145047)

63 or/54-62 (2980563)

64 "filter obs Medline".ti. (0)

65 47 and 63 (12)

66 51 and 63 (52)

67 66 not 65 (40)

68 1 or 2 or 3 (6303)

69 68 and 51 (83)

70 "filter systematic reviews".ti. (0)

71 meta analysis.pt. (98610)

72 (meta-anal$ or metaanal$).af. (169894)

73 (quantitativ$ adj10 (review$ or overview$)).tw. (7826)

74 (systematic$ adj10 (review$ or overview$)).tw. (142782)

75 (methodologic$ adj10 (review$ or overview$)).tw. (10380)

76 (quantitativ$ adj10 (review$ or overview$)).kf. (37)

77 (systematic$ adj10 (review$ or overview$)).kf. (10893)

78 (methodologic$ adj10 (review$ or overview$)).kf. (48)

79 medline.tw. and review.pt. (75023)

80 (pooled adj3 analy\*).tw. (16616)

81 (pooled adj3 analy\*).kf. (170)

82 "cochrane$".fc\_jour. (14838)

83 or/71-82 (301460)

84 "sensitief filter voor RCT's van Cochrane".ti. (0)

85 randomized controlled trial.pt. (516517)

86 controlled clinical trial.pt. (101763)

87 (randomized or randomised).ab. (544119)

88 placebo.ab. (210732)

89 drug therapy.fs. (2200950)

90 randomly.ab. (312542)

91 trial.ab. (478220)

92 groups.ab. (1927737)

93 or/85-92 (4566070)

94 93 not (exp animals/ not humans/) (3951906)

95 34 and 68 (200)

96 95 (200)

97 limit 96 to yr="2000 -Current" (175)

98 97 and (94 or 83) (16)

99 (97 and 63) not (94 or 83) (49)

100 38 and 97 (33)

101 100 not (98 or 99) (16)

102 50 and 97 (86)

**103 102 and (94 or 83) (5) med SR rct**

**104 (102 and 63) not (94 or 83) (26) med observat**

Embase 181

database: Embase <1974 to 2018 January 16>

Search Strategy:

--------------------------------------------------------------------------------

1 "cradock$".fc\_auts. and "language learning".fc\_titl. and "2016".fc\_pubyr. and "1".fc\_pg. (0)

2 "wallace$".fc\_auts. and "single-suture craniosynostosis".fc\_titl. and "2016".fc\_pubyr. (4)

3 from 2 keep 3 (1)

4 craniofacial synostosis/ (6157)

5 craniosynost\*.ti,ab. (4686)

6 craniosynost\*.kw. (1849)

7 4 or 5 or 6 (7147)

8 language/ or language development/ or language processing/ (86253)

9 language delay/ or exp developmental language disorder/ (2373)

10 exp speech/ or "speech and language"/ (84238)

11 (speech or language or speak\*).tw. (252741)

12 (speech or language or speak\*).kw. (26646)

13 or/8-12 (310505)

14 7 and 13 (158)

15 14 (158)

16 limit 15 to yr="2000 -Current" (145)

17 risk/ or risk assessment/ or risk benefit analysis/ or risk factor/ or risk management/ or risk reduction/ (1703803)

18 risk.tw. (2411387)

19 risk.kw. (151247)

20 or/17-19 (2924983)

21 16 and 20 (37) emb risk

22 "filter systematic reviews & meta-analyses Embase".ti. (0)

23 meta analysis/ (136654)

24 "systematic review"/ (153652)

25 (meta-analy$ or metaanaly$).tw. (157534)

26 (systematic$ adj4 (review$ or overview$)).tw. (153171)

27 (quantitativ$ adj5 (review? or overview?)).tw. (4031)

28 (methodologic adj5 (overview? or review?)).tw. (335)

29 (review$ adj3 (database? or medline or embase or cinahl)).tw. (20942)

30 (pooled adj3 analy$).tw. (21825)

31 (extensive adj3 review$ adj3 literature).tw. (3051)

32 (meta or synthesis or (literature adj8 database?) or extraction).tw. (1236482)

33 review.pt. (2318370)

34 32 and 33 (118672)

35 (systematic$ adj4 (review$ or overview$)).kw. (18358)

36 (quantitativ$ adj5 (review? or overview?)).kw. (51)

37 (pooled adj3 analy$).kw. (385)

38 or/23-31,34-37 (408893)

39 "filter rct embase".ti. (0)

40 controlled clinical trial/ or randomized controlled trial/ (657642)

41 randomization/ (76553)

42 Major Clinical Study/ (2975285)

43 random$.tw. (1257125)

44 Double Blind Procedure/ (145234)

45 or/40-44 (4134132)

46 "einde filter rct embase".ti. (0)

47 "filter observationele studies embase".ti. (0)

48 Clinical study/ (154882)

49 Case control study/ (120616)

50 Family study/ (25587)

51 Longitudinal study/ (106730)

52 Retrospective study/ (599970)

53 Prospective study/ (417200)

54 Randomized controlled trials/ (135185)

55 53 not 54 (413050)

56 Cohort analysis/ (334380) 182

57 (Cohort adj (study or studies)).tw. (208449)

58 (Case control adj (study or studies)).tw. (109322)

59 (follow up adj (study or studies)).tw. (56787)

60 (observational adj (study or studies)).tw. (118266)

61 (epidemiologic$ adj (study or studies)).tw. (94422)

62 (cross sectional adj (study or studies)).tw. (153198)

63 or/48-53,55-62 (1961495)

64 "filter observationele studies embase".ti. (0)

65 16 and 38 (10)= emb systrev

66 (16 and (45 or 63)) not 38 (43)= emb rct observat

Cochrane

earch Name: craniosynostoses spraak taal

Last Saved: 17/01/2018 12:28:06.647

Description: gedaan 20180117 HdB

ID Search

#1 MeSH descriptor: [Craniosynostoses] explode all trees

#2 craniosynost\*:ti,ab

#3 craniosynost\*:kw

#4 #1 or #2 or #3

#5 MeSH descriptor: [Language] explode all trees

#6 MeSH descriptor: [Language Disorders] explode all trees

#7 MeSH descriptor: [Speech] explode all trees

#8 MeSH descriptor: [Speech Therapy] explode all trees

#9 MeSH descriptor: [Developmental Disabilities] explode all trees

#10 language or speech or speak\*:ti,ab

#11 #5 or #6 or #7 or #8 or #9 or #10

#12 #4 and #11

database: PsycINFO <1806 to January Week 2 2018>

Search Strategy:

--------------------------------------------------------------------------------

1 craniofacial synostosis.mp. (0)

2 craniosynost\*.ti,ab. (51)

3 craniosynost\*.id. (30)

4 2 or 3 (54)

5 exp LANGUAGE/ or exp LANGUAGE DEVELOPMENT/ or exp LANGUAGE DELAY/ or exp LANGUAGE DISORDERS/ or exp LANGUAGE THERAPY/ (129671)

6 exp oral communication/ or verbal ability/ or verbal fluency/ or exp vocalization/ or exp voice/ (53650)

7 (speech or language or speak\*).tw. (267842)

8 (speech or language or speak\*).id. (115672)

9 or/5-8 (335416)

10 4 and 9 (10)

11 limit 10 to (peer reviewed journal and yr="2000 -Current") (7)

Web of science search strategy with one or more of the previously mentioned terms

|  |  |
| --- | --- |
| **Exclusion after reading full paper**  | **Reasons for exclusion**  |
| Knudsen et al, 2012  | Does not contain relevant outcome measure (see selection- and inclusion criteria)  |
| Knight et al, 2014  | Insufficient quality according to AMSTAR-criteria |
| Toth et al, 2008  | Does not contain outcomes on speach and language, explores whether A not B-tasks are related to PLS-3 amongst others.  |

|  |  |
| --- | --- |
| Judgement of systemic review by Knight et al. (2014) according to AMSTAR **Was an 'a priori' design provided?**  | **No** The authors did not follow the PRISMA and MOOSE guidelines.  |
| **Was there duplicate study selection and data extraction?**  | **Yes/Unclear** Titles and abstracts were screened independently by two reviewers. Unclear how discrepancies were resolved.  |
| **Was a comprehensive literature search performed?**  | **Yes** A literature search was performed across three databases.  |
| **Was the status of publication (i.e. grey literature) used as an inclusion criterion?**  | **Yes** Only peer-reviewed articles were included.  |
| **Was a list of studies (included and excluded) provided?**  | **No** A list of excluded papers is available from the corresponding author on request.  |
| **Were the characteristics of the included studies provided?**  | **Yes**  |
| **Was the scientific quality of the included studies assessed and documented?**  | **Partly** Reviewers did not use validated checklists for assessing risk of bias in case-control studies or cohort studies with a concurrent control group.  |
| **Was the scientific quality of the included studies used appropriately in formulating conclusions?**  | **Unclear**  |
| **Were the methods used to combine the findings of studies appropriate?**  | **Studies were not combined due to heterogeneity**  |
| **Was the likelihood of publication bias assessed?**  | **Yes** It is acknowledged that limiting inclusion to peer-reviewed published studies in English language may have introduced a potential publication bias.  |
| **Was the conflict of interest included?**  | **No**  |