BTS Guideline for diagnosing and monitoring paediatric sleep disordered breathing

Online Appendix 4 Question 4 Evidence Review and Protocol

Q4 What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

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Question Evidence Review

Q4 What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

Background

There are particular comorbid disorders seen in children that predispose to sleep disordered breathing (SDB). Children with comorbidities may have less obvious symptoms of SDB than typically developing children and therefore screening is often advocated. Current tests in the UK for diagnosing or detecting SDB in children are pulse oximetry, cardiorespiratory sleep study (CRSS) and polysomnography (PSG). When choosing which test to perform, considerations include which tests are accessible to the entire UK paediatric population, which tests are cost effective and which tests can accurately identify SDB. In the UK there is a desire to use simple investigations to detect SDB wherever possible, so this review evaluates the diagnostic accuracy of pulse oximetry and CRSS to diagnose SDB in children with comorbid disorders predisposing to SDB.

Outcomes

Diagnostic accuracy of pulse oximetry and CRSS to diagnose SDB in children with comorbid disorders predisposing to SDB

Evidence Review

The initial literature search identified 201 potentially relevant papers, but only six were deemed suitable for the review.¹⁻⁶ All studies that did not specifically address the review question or were not truly reflective of standard UK pulse oximetry or CRSS clinical practice were excluded from the review. All studies used PSG as a gold standard. Only a limited number of comorbid conditions were identified in these papers (Robin sequence¹, Down Syndrome³, neuromuscular disease⁵, myelomeningocele⁴ or varied disorders²), which is likely to be a source of bias and heterogeneity, and study groups included asymptomatic and symptomatic children. Due to the limited number of relevant studies, there was also heterogeneity in the pulse oximetry and CRSS parameters used across the studies (<u>Table 4a</u>).

	Table 4a: Pulse	oximetry	and CRSS	parameters
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PSG cut-off	Oximetry/CRSS variable(s) measured/cut-off
AHI >10	Minimum SpO ₂ <80%
MOAHI >1	≥3 clusters of 3% SpO₂ dips to <90%
OAHI ≥1	McGill score (cut-off not specified)
AHI ≥5	'Moderate-to-severe' SBD from oximetry (exact definition not specified)
AHI >1	AHI>1
AHI ≥5	AHI ≥5
AHI ≥1	AHI≥1
	AHI >10 MOAHI >1 OAHI ≥1 AHI ≥5 AHI >1 AHI ≥5

AHI – apnoea hypopnoea index; MOAHI – mixed/obstructive apnoea hypopnoea index; OAHI – obstructive apnoea hypopnoea index; SDB – sleep disordered breathing

* Studies used a mix of PSG or CRSS as a gold standard, but were included in the meta-analyses because to the lack of supporting evidence

Pulse Oximetry

Four studies investigated the diagnostic accuracy of pulse oximetry in the diagnosis of SDB in children with comorbid disorders. Meta-analysis of all available data, regardless of AHI cut-off, showed a sensitivity and specificity of 0.49 [0.31 to 0.67] and 0.87 [0.78 to 0.93] respectively [95% confidence intervals] (Figure 4a).¹⁻⁴

Pulse Oximetry (AHI ≥1)

Sub-analysis of the diagnostic accuracy of pulse oximetry to diagnose an apnoea-hypopnea index (AHI) \geq 1 in children with comorbid disorders gave a pooled estimate sensitivity of 0.43 [0.32 to 0.54] and specificity of 0.93 [0.84 to 0.98] [95% confidence intervals] (Figure 4b).^{2,3}

CRSS

Three analyses evaluated the diagnostic accuracy of CRSS for diagnosing SDB in children with comorbid disorders. Meta-analysis of the results showed a pooled sensitivity of 0.76 [0.47, 0.92] and pooled specificity of 0.62 [0.24, 0.89] [95% confidence intervals] (Figure 4c).^{5,6}

CRSS (AHI ≥1)

Two studies specifically investigated the diagnostic accuracy of CRSS for diagnosing AHI \geq 1, reporting a sensitivity and specificity of 0.85 [0.35, 0.98] and 0.41 [0.13, 0.76] respectively [95% confidence intervals] (Figure 4d).^{5,6}

CRSS (AHI ≥5)

One study investigated the diagnostic accuracy of CRSS for diagnosing AHI \geq 5, reporting a sensitivity and specificity of 0.62 [0.32, 0.86] and 0.87 [0.60, 0.98] respectively [95% confidence intervals].⁵

A summary of the pulse oximetry and CRSS results is shown in <u>Table 4b</u>.

Table 4b: Diagnostic accuracies of pulse oximetry and cardiorespiratory sleep study for diagnosing sleep disordered breathing in children

Included data	No. of datasets	Sensitivity [95% CI]	Specificity [95% CI]
Pulse oximetry (all)	4	0.49 [0.31, 0.67]	0.87 [0.78, 0.93]
<u>Pulse oximetry (AHI ≥1)</u>	2	0.43 [0.32, 0.54]	0.93 [0.84, 0.98]
<u>CRSS (all)</u>	3	0.76 [0.47, 0.92]	0.62 [0.24, 0.89]
<u>CRSS (AHI ≥1)</u>	2	0.85 [0.35, 0.98]	0.41 [0.13, 0.76]
CRSS (AHI ≥5)⁵	1	0.62 [0.32, 0.86]	0.87 [0.60, 0.98]

CI – confidence intervals; CRSS – cardiorespiratory sleep study

Evidence statements

Pulse oximetry appears to have a low sensitivity and high specificity for diagnosing sleep disordered breathing in children with comorbid disorders (<u>Very Low</u>)

Pulse oximetry appears to have a low sensitivity and very high specificity for diagnosing mild-to-moderate sleep disordered breathing in children with comorbid disorders (<u>Very Low</u>)

Cardiorespiratory sleep studies appear to have a moderate sensitivity and low specificity for the diagnosis of sleep disordered breathing in children with neuromuscular disorders and Down Syndrome (<u>Very Low</u>)

Recommendation

 For children with neuromuscular disorders or Down Syndrome predisposing to sleep disordered breathing, cardiorespiratory sleep studies (CRSS) can be considered for diagnosing sleep disordered breathing (<u>Conditional</u>)

Good Practice Point

- ✓ Although CRSS can only be recommended as a diagnostic tool for sleep disordered breathing in children with neuromuscular disorders or Down Syndrome, CRSS can be considered as a first line diagnostic tool for children with other comorbidities
- ✓ If a CRSS is abnormal, the significance of the findings should be carefully considered and the range of potential management options discussed with the child and their family/carer
- ✓ If CRSS findings are inconsistent with the clinical picture, the clinical history should be reviewed giving specific consideration to non-respiratory causes of sleep disorders. Referral to a neurology sleep service for assessment should also be considered
- ✓ If a CRSS is not available pulse oximetry can be considered as an initial diagnostic test for sleep disordered breathing in children with comorbid disorders, but if a test result is abnormal caution must be taken in interpreting the results as desaturations may have varying causes. Referral for more complex studies may be required to assess for hypoventilation and determine the cause and mechanisms of desaturation
- ✓ If a pulse oximetry test is normal this does not exclude sleep disordered breathing and clinical review should consider repeat/additional testing
- ✓ As desaturations are non-specific, if considering pulse oximetry for diagnosing sleep disordered breathing in children with comorbidities caution must be taken when interpreting the results
- ✓ If CRSS or pulse oximetry is inappropriate, or if a CRSS or pulse oximetry test result is inconsistent with the clinical picture, and non-respiratory causes of sleep disorders have been ruled out, then polysomnography (PSG) should be considered
- ✓ Clinicians are cautioned from using AHI alone to guide decision making
- ✓ If hypoventilation is suspected, guideline users should refer to Supplementary Online Appendix 3

Research Recommendation

 Research is needed into determining the diagnostic accuracy of pulse oximetry and cardiorespiratory sleep studies as a screening tool for diagnosing sleep disordered breathing in children with comorbid disorders, including cerebral palsy, Down Syndrome, neuromuscular disorders, craniofacial disorders and storage disorders

Meta-analyses

Diagnostic accuracy table contents and summary receiver operating characteristic (SROC) curve legend

Table contents

Pooled sensitivity [95% confidence intervals]

Pooled specificity [95% confidence intervals]

Likelihood ratio of a positive test result (LR+) [95% confidence intervals]

Likelihood ratio of a negative test result (LR-) [95% confidence intervals]

Diagnostic odds ratio (DOR, an indicator of the likelihood of a positive test result) [95% confidence intervals]

Summary receiver operating characteristic (SROC) curve legend

– SROC

- Study estimate
- Summary point
- --- 95% prediction region

Figure 4a Pulse oximetry (all data)

Study	TP	FP	FN	TN	Sensitivity (95% CI)	Specificity (95% CI)	Sensitivity (95% CI)	Specificity (95% CI)
Aaronson 2017	5	8	11	22	0.31 [0.11, 0.59]	0.73 [0.54, 0.88]		
Brouilete 2000	19	3	26	44	0.42 [0.28, 0.58]	0.94 [0.82, 0.99]		
Lin 2014	15	1	20	13	0.43 [0.26, 0.61]	0.93 [0.66, 1.00]		
Waters 1998	14	8	3	58	0.82 [0.57, 0.96]	0.88 [0.78, 0.95]		



r colou conolavity	0.100	[0.000, 0.007]
Pooled Specificity	0.873	[0.780, 0.930]
LR+	3.816	[0.911, 6.721]
LR-	0.590	[0.359, 0.821]
DOR	6.472	[-0.670, 13.614]

Figure 4b Pulse oximetry (AHI ≥1)

Study	TP	FP	FN	TN	Sensitivity (95% CI)	Specificity (95% CI)	Sensitivity (95% CI)	Specificity (95% CI)
Brouilete 2000	19	3	26	44	0.42 [0.28, 0.58]	0.94 [0.82, 0.99]		
Lin 2014	15	1	20	13	0.43 [0.26, 0.61]	0.93 [0.66, 1.00]	0 0.2 0.4 0.6 0.8 1	

Pooled Sensitivity	0.425	[0.322, 0.535]
Pooled Specificity	0.934	[0.838, 0.975]
LR+	6.481	[0.123, 12.839]
LR-	0.615	[0.492, 0.738]
DOR	10.533	[-1.121, 22.187]

Figure 4c CRSS (all data)

Study	TP	FP	FN	TN	Sensitivity (95% CI)	Specificity (95% Cl) Sensitivity (95% CI)	Specificity (95% CI)
Fishman 2018 AHI1	15	2	7	4	0.68 [0.45, 0.86]	0.67 [0.22, 0.96]	
Fishman 2018 AHI5	8	2	5	13	0.62 [0.32, 0.86]	0.87 [0.60, 0.98]	
lkizoglu 2019 AHI1	6	10	0	3	1.00 [0.54, 1.00]	0.23 [0.05, 0.54		
							0 0.2 0.4 0.6 0.8 1	0 0.2 0.4 0.6 0.8 1
Pooled Sensitivity	y	C).76	2	[0.474,	0.919]		
Pooled Specificity	y	C	0.61	6	[0.242,	0.889]		
LR+		1	1.98	2	[0 834	4.713]		
			1.50	2	[0.004,	4.710]		
LR-		C).38	6	[0.170,	0.877]		
DOR		5	5.13	0	[1.251,	21.037]		

Figure 4d CRSS (AHI ≥1)

Study Fishman 2018 Ikizoglu 2019	TP 15 6	FP 2 10	FN 7 0	TN 4 3	Sensitivity (95% Cl) 0.68 [0.45, 0.86] 1.00 [0.54, 1.00]	Specificity (95% Cl) 0.67 [0.22, 0.96] 0.23 [0.05, 0.54]	Sensitivity (95% Cl)	Specificity (95% Cl)
Pooled Sens	itivity	/	C	.848	3 [0.35	3, 0.983]		
Pooled Spec	ificity	/	С	.413	3 [0.13	4, 0.762]		
LR+			1	.444	4 [0.80	1, 2.604]		
LR-			C	.386	6 [0.05	9, 2.286]		
DOR			3	8.92 <i>°</i>	1 [0.41	9, 36.697]		

Risk of bias summary

		Risk (of Bias	5	Applicability Concerns					
	Patient Selection	Index Test	Reference Standard	Flow and Timing	Patient Selection	Index Test	Reference Standard			
Aaronson 2017	•	?	?	•	•	?	•			
Brouillette 2000	•	•	•	•	•	•	•			
Fishman 2018	•	•	•	•	•	•	•			
lkizoglu 2019	•	?	?	•	•	?	•			
Lin 2014	•	?	?	•	•	?	•			
Waters 1998	•	•	•	•	•	•	•			
e High			<mark>?</mark> Uı	ıclear			+ Lo	w		

GRADE analyses

Pulse oximetry (all data)

What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

Patient or population: Children (<17 years) with comorbid disorders predisposing to sleep disordered breathing New test: Pulse oximetry

Pooled sensitivity: 0.48 (95% CI: 0.31 to 0.67) | Pooled specificity: 0.87 (95% CI: 0.78 to 0.93)

Test result	Number of results per 1,000 patients tested (95% CI)	Number of participants (studies)	Certainty of the Evidence (GRADE)	
	Prevalence 40%* Typically seen in			
True positives	194 (123 to 267)	113	$\oplus 000$	
False negatives	206 (133 to 277)	(4)	VERY LOW a,b	
True negatives	524 (468 to 558)	157	$\oplus 000$	
False positives	76 (42 to 132)	(4)	VERY LOW ^{a,b}	
	Prevalence 60%*			
	Typically seen in			
True positives	291 (185 to 400)	113	000	
False negatives	309 (200 to 415)	(4)	VERY LOW ^{a,b}	
True negatives	349 (123 to 372)	157	$\oplus 000$	
False positives	51 (28 to 88)	(4)	VERY LOW ^{a,b}	
	Prevalence 80 %* Typically seen in			
True positives	388 (246 to 534)	113	000	
False negatives	412 (266 to 554)	(4)	VERY LOW ^{a,b}	
True negatives	175 (156 to 186)	157	$\oplus 000$	
False positives	25 (14 to 44)	(4)	VERY LOW ^{a,b}	
CI: Confidence interval				

Explanations

a. High risk of bias across studies

b. Sensitivity inconsistency in one study

Pulse oximetry (AHI ≥1)

What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

Patient or population: Children (<17 years) with comorbid disorders predisposing to sleep disordered breathing New test: Pulse oximetry (AHI ≥1)

Pooled sensitivity: 0.42 (95% CI: 0.32 to 0.54) | Pooled specificity: 0.95 (95% CI: 0.84 to 0.97)

Test result	est result Number of results per 1,000 patients tested (95% CI)		Certainty of the Evidence (GRADE)
	Prevalence 40%* Typically seen in		
True positives	170 (129 to 214)	80	$\oplus 000$
False negatives	230 (186 to 271)	(2)	VERY LOW ^{a,b}
True negatives	560 (503 to 585)	61	000
False positives	40 (15 to 97)	(2)	VERY LOW ^{a,b}
	Prevalence 60 %* Typically seen in		
True positives	255 (193 to 321)	80	⊕000
False negatives	345 (279 to 407)	(2)	VERY LOW ^{a,b}
True negatives	374 (355 to 390)	61	000
False positives	26 (10 to 65)	(2)	VERY LOW ^{a,b}
	Prevalence 80%* Typically seen in		
True positives	340 (258 to 428)	80	$\oplus 000$
False negatives	460 (372 to 542)	(2)	VERY LOW ^{a,b}
True negatives	187 (168 to 195)	61	000
False positives	13 (5 to 32)	(2)	VERY LOW a,b
CI: Confidence interval			
Explanations			

Explanations

a. High risk of bias across studies

b. GRADE lowered by one score as result based on two studies

CRSS (all data)

What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

Patient or population: Children (<17 years) with comorbid disorders predisposing to sleep disordered breathing New test: CRSS

Pooled sensitivity: 0.76 (95% CI: 0.47 to 0.92) | Pooled specificity: 0.62 (95% CI: 0.24 to 0.89)

Test result	est result Number of results per 1,000 patients tested (95% CI)		Certainty of the Evidence (GRADE)
	Prevalence 40%* Typically seen in		
True positives	305 (190 to 368)	41	000
False negatives	95 (32 to 210)	(3)	VERY LOW a,b,c
True negatives	370 (145 to 533)	34	$\oplus 000$
False positives	230 (67 to 455)	(3)	VERY LOW a,b,c
	Prevalence 60 %* Typically seen in		
True positives	457 (284 to 551)	41	000
False negatives	143 (49 to 316)	(3)	VERY LOW a,b,c
True negatives	246 (97 to 356)	34	$\oplus 000$
False positives	154 (44 to 303)	(3)	VERY LOW a,b,c
	Prevalence 80%* Typically seen in		
True positives	610 (379 to 735)	41	0000
False negatives	190 (65 to 421)	(3)	VERY LOW a,b,c
True negatives	123 (48 to 178)	34	$\oplus 000$
False positives	77 (22 to 152)	(3)	VERY LOW a,b,c
CI: Confidence interval			
Explanations			

a. Some risk of bias across studies

b. Serious inconsistency across the studies

c. Moderate confidence intervals in some datasets

CRSS (AHI ≥1)

What is the diagnostic accuracy of pulse oximetry versus cardiorespiratory sleep studies for children with comorbid disorders predisposing to sleep disordered breathing?

Patient or population: Children (<17 years) with comorbid disorders predisposing to sleep disordered breathing New test: CRSS (AHI ≥1)

Pooled sensitivity: 0.85 (95% CI: 0.35 to 0.98) | **Pooled specificity**: 0.41 (95% CI: 0.13 to 0.76)

Test result	Number of results per 1,000 patients tested (95% Cl)	Number of participants (studies)	Certainty of the Evidence (GRADE)
	Prevalence 40%* Typically seen in		
True positives	339 (141 to 393)	222	⊕ 000
False negatives	61 (7 to 259)	(9)	VERY LOW a,b,c,d
True negatives	248 (80 to 457)	667	$\oplus 000$
False positives	352 (143 to 520)	(9)	VERY LOW a,b,c,d
	Prevalence 60 %* Typically seen in		
True positives	509 (212 to 590)	222	000
False negatives	91 (10 to 388)	(9)	VERY LOW a,b,c,d
True negatives	165 (54 to 305)	667	000
False positives	235 (95 to 346)	(9)	VERY LOW a,b,c,d
	Prevalence 80%* Typically seen in		
True positives	678 (282 to 786)	222	0000
False negatives	122 (14 to 518)	(9)	VERY LOW a,b,c,d
True negatives	83 (27 to 152)	667	$\oplus 000$
False positives	117 (48 to 173)	(9)	VERY LOW a,b,c,d
CI: Confidence interval			
Explanations			

a. Some risk of bias across studies

b. Some indirectness - 2 different disease conditions

c. Inconsistency across the studies

d. Moderate confidence intervals

Recommendation Tables

Question Details

POPULATION:	Children (<17 years) with suspected sleep disordered breathing and comorbid disorder(s)
INDEX TESTS:	Pulse oximetry and cardiorespiratory sleep study (CRSS)
GOLD STANDARD:	Polysomnography (PSG)
OUTCOME:	Diagnostic accuracy of CRSS for diagnosing sleep disordered breathing in infants and children with comorbid disorders

Pulse oximetry

SUMMARY OF JUDGEMENTS

			JU	DGEMENT			
PROBLEM	No	Probably no	Probably yes	Yes		Varies	Don't know
TEST ACCURACY	Very inaccurate	Inaccurate	Accurate	Very accurate		Varies	Don't know
DESIRABLE EFFECTS	Trivial	Small	Moderate	Large		Varies	Don't know
UNDESIRABLE EFFECTS	Large	Moderate	Small	Trivial		Varies	Don't know
CERTAINTY OF EVIDENCE	Very low	Low	Moderate	High			No included studies
BALANCE OF EFFECTS	Favours the comparison	Probably favours the comparison	Does not favour the intervention or the comparison	Probably favours the intervention	Favours the intervention	Varies	Don't know

TYPE OF RECOMMENDATION

Strong recommendation against the intervention	Conditional recommendation against the intervention	Conditional recommendation for either the intervention or the comparison	Conditional recommendation for the intervention	Strong recommendation for the intervention
	\boxtimes			

CONCLUSIONS

Recommendation

Currently pulse oximetry should not be used for diagnosing sleep disordered breathing in children with comorbid disorders

Justification

Pulse oximetry has a low sensitivity and high specificity for diagnosing sleep disordered breathing in children with comorbid disorders (<u>Very Low</u>)

Subgroup considerations

There were not enough data to consider children with cerebral palsy, Down Syndrome, craniofacial disorders, or storage disorders

Research priorities

Research is needed into determining the diagnostic accuracy of pulse oximetry as a screening tool for diagnosing sleep disordered breathing in children with comorbid disorders, including cerebral palsy, Down Syndrome, neuromuscular disorders, craniofacial disorders and storage disorders

CRSS

SUMMARY OF JUDGEMENTS

			JL	IDGEMENT			
PROBLEM	No	Probably no	Probably yes	Yes		Varies	Don't know
TEST ACCURACY	Very inaccurate	Inaccurate	Accurate	Very accurate		Varies	Don't know
DESIRABLE EFFECTS	Trivial	Small	Moderate	Large		Varies	Don't know
UNDESIRABLE EFFECTS	Large	Moderate	Small	Trivial		Varies	Don't know
CERTAINTY OF EVIDENCE	Very low	Low	Moderate	High			No included studies
BALANCE OF EFFECTS	Favours the comparison	Probably favours the comparison	Does not favour the intervention or the comparison	Probably favours the intervention	Favours the intervention	Varies	Don't know

TYPE OF RECOMMENDATION

Strong recommendation against the intervention	Conditional recommendation against the intervention	Conditional recommendation for either the intervention or the comparison	Conditional recommendation for the intervention	Strong recommendation for the intervention
			\boxtimes	

CONCLUSIONS

Recommendation

For children with neuromuscular disorders predisposing to sleep disordered breathing, cardiorespiratory sleep studies can be considered for diagnosing sleep disordered breathing

Justification

Cardiorespiratory sleep studies have a moderate sensitivity and low specificity for the diagnosis of sleep disordered breathing in children with neuromuscular disorders and Down Syndrome (<u>Very Low</u>)

Subgroup considerations

There were not enough data to consider children with cerebral palsy, Down Syndrome, craniofacial disorders, or storage disorders

Research priorities

Research is needed into determining the diagnostic accuracy of cardiorespiratory sleep studies as a screening tool for diagnosing sleep disordered breathing in children with comorbid disorders, including cerebral palsy, Down Syndrome, neuromuscular disorders, craniofacial disorders and storage disorders

References

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Question Protocol

Field	Content
Review Question	For asymptomatic children with comorbid disorders and predisposing to sleep disordered breathing, what is the diagnostic accuracy of pulse oximetry and cardiorespiratory sleep studies?
Type of review question	Diagnostic accuracy
Objective of the review	Certain groups of children (e.g. those with neuro-disability) may have less obvious symptoms of sleep disordered breathing than typically developing children and therefore screening is advocated. It is uncertain what test modality is most appropriate. Oximetry is much simpler and less obtrusive than cardiorespiratory sleep studies and can be done at home. This may be particularly important for this group of children.
	 Does normal oximetry rule out significant SDB? How sensitive and specific is oximetry? Is there a difference between home and hospital oximetry?
Eligibility criteria – population / disease / condition / issue / domain	Children (<17 years) with comorbid disorders predisposing to sleep disordered breathing
Eligibility criteria – index test(s)	Pulse oximetry Cardiorespiratory sleep studies
Eligibility criteria – gold standard	Polysomnography
Outcomes and prioritisation	Diagnostic accuracy
Eligibility criteria – study design	Randomised controlled trials Observational studies Superiority studies Case series
Other inclusion /exclusion criteria	Non-English language excluded unless full English translation Conference abstracts, Cochrane reviews, systematic reviews, reviews Cochrane reviews and systematic reviews can be referenced in the text, but DO NOT use in a meta-analysis

Proposed sensitivity / subgroup analysis, or meta- regression	Children <2 years with cerebral palsy Children 2-16 years with cerebral palsy Children <2 years with Down Syndrome Children 2-16 years with Down Syndrome Children <2 years with craniofacial disorders Children 2-16 years with craniofacial disorders Children <2 years with storage disorders Children 2-16 years with storage disorders
Selection process – duplicate screening / selection / analysis	Agreement should be reached between Guideline members who are working on the question. If no agreement can be reached, a decision should be made by the Guideline co-chairs. If there is still no decision, the matter should be brought to the Guideline group and a decision will be made by consensus
Data management (software)	RevMan5Meta-analysis data input. Evidence review/considered judgement. Storing Guideline text, tables, figures, etc.MetaDTAData meta-analysesGradeproQuality of evidence assessment / Recommendations
Information sources – databases and dates	MEDLINE, Embase, PubMED, Central Register of Controlled Trials and Cochrane Database of Systematic Reviews No date restriction
Methods for assessing bias at outcome / study level	RevMan5 diagnostic accuracy full review template (based on QUADAS2) (follow instructions in ' <i>BTS Guideline Process Handbook - Diagnostic Accuracy</i> ')
Methods for quantitative analysis – combining studies and exploring (in)consistency	If 3 or more relevant studies: RevMan5 for forest plots, summary ROC plot MetaDTA to combine studies (pooled specificity, sensitivity, likelihood ratios, diagnostic odds ratio and confidence intervals) and calculate RevMan parameters for summary ROC plot (follow instructions in <i>'BTS Guideline Process Handbook - Diagnostic</i> <i>Accuracy'</i>)
Meta-bias assessment – publication bias, selective reporting bias	 GRADEpro Diagnostic accuracy quality of evidence assessment for each index test (follow instructions in '<i>BTS Guideline Process Handbook - Diagnostic Accuracy</i>')

Rationale / context – what is	There is very little known about simpler testing modalities such as oximetry
known	in high-risk children. The literature contains some cross-sectional studies in
	those with Down Syndrome and craniofacial disorders.