Table 9: Studies included in monitoring respiratory health in adults with cerebral palsy

Bibliographic details	Number of Participant & Participant Characteristics	Test/Outcome characteristics	Outcome measures to be used	Results	Reviewer comment
Authors Lampe, R., Blumenstein, T., Turova, V., Alves-Pinto, A. **Year of publication** 2014 **Country of publication** Germany **Ref Id** 347233 **Consecutive recruitment** Not reported	**Cohort population** Adults with cerebral palsy (CP) were recruited from a general rehabilitation and training program at a centre for persons with CP. **Inclusion Criteria** Not reported. **Exclusion Criteria** Not reported **Demographics - Total** 46 **Cases** 8 (with OS < 96%) **Statistical method** Vital capacity (VC) was measured with with a spirometer (MiniSpir ®) from MIR (Medical International Research Srl, Rome, Italy). The spirometer was connected to a computer which recorded the measurements. Each participant was asked to completely enclose	**Reference Test** A pulse oximeter (PO80; Beurer GmbH, Ulm, Germany) clamped to the tip of the finger was used to measure the heart rate and OS in the blood. Readings were taken directly from the device or recorded on a computer via a USB connection. Recordings were made over a period of 5 minutes at a sampling rate of 1 Hz. The average value over the measurement period was calculated.	**Raw Data** See table 1 in Lampe (2014)	Using deviation from normal vital capacity (DVC) thresholds to predict abnormally low oxygen saturation (<96%). Calculated from Table 1 in Lampe (2014)[1] **DVC, TP, FP, FN, TN, Sn [95%CI], Sp [95%CI],** -2.0, 7, 31, 0, 0,1.00 [0.59, 1.00], 0.00 [0.00, 0.11] -0.5, 7, 30, 0, 4,1.00 [0.59, 1.00],0.12 [0.03, 0.27] 0.0, 7,28,0,3,1.00 [0.59, 1.00], 0.10 [0.02, 0.26] 0.2, 7,27, 0,4,1.00 [0.59, 1.00], 0.13 [0.04, 0.30] 1.0, 6,23,1,8, 0.86 [0.42, 1.00],0.26 [0.12, 0.45] 1.5,5,16,2,15,0.71 [0.29, 0.96],0.48 [0.30, 0.67] 2.0,3,14,4,17,0.43 [0.10, 0.82],0.55 [0.36, 0.73] 2.5,2,4,5,27,0.29 [0.04, 0.71],0.87 [0.70, 0.96] 3.0,0,1,7,30,0.00 [0.00, 0.41],0.97 [0.83, 1.00] 4.0,0,0,7,31,0.00 [0.00, 0.41],1.00 [0.89, 1.00] There was a significant positive correlation between lung vital capacity and chest expansion (P<0.05). But there was no statistically significant correlation between lung vital capacity and oxygen	**Funding** Not reported (authors report no conflicts of interest). **Quality Items** QUADAS 2 checklist Patient selection Risk of bias: Was a consecutive or random sample of patients enrolled? Unclear Was a case-control design avoided? Yes Did the study avoid inappropriate exclusions? Unclear Could the selection of participants have introduced bias? Unclear

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	the turbine with his or her mouth, to inhale up to his or her maximum capacity, and then to exhale with the maximum force possible. Some people could not enclose the turbine completely with their mouths. In these cases, a mouthpiece made of rubber was used. The VC was the difference in the volume between maximal inhalation and maximum exhalation. The measurement was repeated four to five times, and the maximum value used. Chest expansion (CE) was measured with a measuring tape at maximum inspiration and expiration, and during normal breathing. The tape was slightly below the mammillary line. CE was calculated as the difference, in cm, between the perimeter of the chest at maximum inspiration and that at full expiration. The normal values of VC were calculated using the following formulas: VC (male, in l) = 10 -3 × (27.63 - (0.112× age)) × height (cm)			saturation, or between chest expansion and oxygen saturation. As GMFCS level increased the mean lung vital capacity and the chest expansion decreased. Oxygen saturation was typically within the normal range, in spite of reduced lung vital capacity and chest expansion. Although scoliosis was associated with an additional decrease in lung vital capacity, this did not affect blood oxygen supply. So although there was decreased chest expansion and the significantly reduced lung volume in these adults with cerebral palsy, there appeared to be sufficient oxygen supply.	Applicability: Is there concern that the included participants do not match the review question? Unclear Index tests Risk of bias: Were the index tests interpreted without knowledge of the reference standard? Unclear If a threshold was used, was it pre-specified? Not applicable (thresholds were applied by the reviewer) Could the conduct or interpretation of the index test have introduced bias? Unclear Applicability: Is there concern that the index test, its conduct or interpretation differ

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	VC (female, in l) = 10 -3 × (21.78 - (0.101× age)) × height (cm) This was used to calculate the deviation of VC from the normal value (DVC) **Diagnostic criteria** Normal oxygen saturation (OS) was defined as 96% to 100%.				from the review question? Unclear Reference standard Risk of bias: Is the reference standard likely to correctly classify the target condition? Yes Were the reference standard results interpreted without knowledge of the results of the index test? Yes Could the reference standard, its conduct or interpretation have introduced bias? No Applicability: Is there concern that the target condition as defined by the reference standard does not match the review question? No Flow and timing Risk of bias: Was there an appropriate interval

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					between index tests and reference standard? Not reported Did all participants receive a reference standard? Yes Did participants receive the same reference standard? Yes Were all patients included in the analysis? Yes (some tests were not possible - but these are accounted for) Could the participant flow have introduced bias? No **OVERALL ASSESSMENT:** Very low quality
Authors Lennon, N., Thorpe, D., Balemans, A. C., Fragala-	**Cohort population** 117 teenagers or adults with cerebral palsy (ages ranged from 16 to 67), reported in 7 studies.	**Reference Test** Aerobic capacity: • VO2 plateau • Heart rate		**Results** **Bicycle ergometer** 4 studies (N=101), age (16 to 67), criterion validity not applicable, construct	Funding Grant (Fit Active Habits) from the American Academy of Cerebral Palsy

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Pinkham, M., O'Neil, M., Bjornson, K., Boyd, R., Dallmeijer, A. J. **Year of publication** 2015 **Country of publication** USA / Netherlands Ref Id 545935 **Consecutive recruitment** Not applicable **Sub-type** Systematic review	All were ambulatory or wheelchair users. GMFCS I (N=45), II (N=25), not reported (N=57) 3 studies (N=42) included only athletes. **Inclusion Criteria** Studies were included if they met the following criteria: • evaluated clinimetric properties of lab or field-based aerobic or anaerobic fitness capacity measures • had a study population who had a diagnosis of CP, • were specific to adolescents (14–18 yrs) and/or adults (>18 yrs) - at least 75% of included participants had to meet the age criteria • published as full reports. **Exclusion Criteria** Studies were excluded if: • they were not published in English, • maximal aerobic exercise test protocols were not	• Respiratory exchange ratio		validity moderate, test-retest reliability moderate, measurement error moderate **Wheelchair ergometer** 3 studies (N=22), age (18 to 33), criterion validity not applicable, construct validity unknown, test-retest reliability limited, measurement error unknown **6 minute walk test** 1 study (N=41), age (16 to 24), criterion validity strong, construct validity not reported, test-retest reliability not applicable, measurement error not applicable	and Developmental Medicine (AACPDM). Quality Items CASP systematic review checklist: Did the review address a clearly focused question? Yes Did the review include the right type of study? Unclear (the included studies do not report clinical outcomes) Did the reviewers try to identify all relevant studies? Yes Did the reviewers do enough to assess the quality of the included studies? Yes If the results of the studies have been combined, was it reasonable to do so? Yes (results

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	incremental or only measured ability/activity/efficiency, • they used self-report measures • they were review articles, single case studies, or commentaries. **Demographics - Total** 117 **Cases** Not applicable **Statistical method** The overall level of evidence for aerobic fitness tests was derived by combining the results of the methodological quality ranking for the studies with the statistical findings for each clinimetric property (validity & reliability). The Cochrane Back Review Group levels of evidence were used (van Tulder et al., 1997). • strong (consistent findings in multiple studies of good methodological quality or in one study of excellent methodological quality) • moderate (consistent findings in multiple studies of fair methodological quality or in				have been combined for similar tests) How are the results presented and what is the main result? (see results section) How precise are these results? Unclear (there is no pooled effect estimate) Can the results be applied to the local population? Unclear (all ambulatory or self-propelled wheelchair users, high proportion were athletes) Are the benefits worth the harms and costs? Unclear (clinical outcomes not reported) Overall this systematic review provides only indirect evidence about the impact of respiratory

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	one study of good methodological quality) • limited (one study of fair methodological quality); • conflicting (conflicting findings); • Unknown (only studies of poor methodological quality). **Diagnostic criteria** Lab based aerobic capacity tests: • Bicycle ergometer • Wheelchair ergometer Field based aerobic capacity tests: • 6 minute walk test				monitoring on patient outcomes - (very low quality) Other information – methodological quality of the individual studies in this review was appraised by the review authors using the COSMIN checklist.

AACPDM: American Academy of Cerebral Palsy and Developmental Medicine; CASP: Critical appraisal skills programme; CE: chest expansion; COSMIN: Consensus-based Standards for the Selection of Health Status Measurement Instruments; CP: cerebral palsy; DVC: Direct vital capacity; GMFCS: Gross motor function classification system; MIR: Medical International Research; NGA: National Guidelines Alliance; OS: Oxygen saturation; QUADAS: Quality Assessment of Diagnostic Accuracy Studies VC: Vital capacity
1. Calculated by the NGA team