Online Table S2.1: Patients (pathologies) who may benefit from continuous positive airway pressure (CPAP).

Author	Country	Journal	Type of study	Number of patients	Ages	Pathologies	Comments
Afsharpaiman et al. [1]	Australia	Sleep & Breathing	Retrospective cohort	46 children 25 had OSA	3 m-14 yrs	Achondroplasia	9 treated with CPAP = 9.8% of those > 2 yrs and 28% of those < 2 yrs
Mogayzel et al. [2]	USA	J Pediatr	Prospective cohort	88 children	1 m-12.6 yrs	Achondroplasia	2 treated with CPAP
Schluter et al. [3]	Germany	Georgian Med News	Cohort study	22 + 2 children	1.3 m-15 yrs	22 achondroplasia + 2 hypochondroplasia	3 treated with CPAP or BPAP
Waters et al. [4]	Australia	Am J Med Gen	Cohort study	30 children	1-47.6 yrs	Achondroplasia	13 treated with CPAP
Julliand et al. [5]	France	Am J Med Gen	Retrospective cohort	30 children	0.4-17.1 yrs	Achondroplasia	1 treated with CPAP, 4 with BPAP
Tenconi et al. [6]	France	Am J Med Gen	Retrospective cohort	43 children	0.3-13.3 yrs	Achondroplasia	2 treated with CPAP
Fauroux et al. [7]	France	AJRCCM	Retrospective cohort	12 children	8 m-6.5 yrs	Laryngomalacia	12 treated with CPAP
Zwacka et al. [8]	Germany	Sleep & Breathing	Retrospective cohort	10 infants	1 to 5 m	Laryngomalacia	10 treated with CPAP
Essouri et al. [9]	France	Intensive Care Med	Prospective physiological + clinical study (Poeso + Pgas measures)	10 infants	3-18 m	5 laryngomalacia (1 + Down syndrome), 3 tracheomalacia (1 + Down syndrome), 1 tracheal hypoplasia, 1 Pierre Robin Sequence	BPAP associated with patient-ventilator asynchrony (trigger insufficiently sensitive)
Kawaguchi et al. [10]	USA	J Pediatr Surgery	Retrospective cohort	4 type III and 5 type IV	8 m-21 yrs	Tracheomalacia after laryngotracheal cleft	5 treated with CPAP

				laryngotracheal cleft		repair	
Pellen et al. [11]	Australia	Int J Pediatr Otorhinolaryngology	Retrospective cohort	16 infants (1 death)	0-9 m	Congenital tracheal stenosis	10/15 treated with CPAP
Shatz et al. [12]	Israel	Otol Rhinol Laryngol	Retrospective cohort	50 infants	1-18 m	Pharyngomalacia	9/50 treated with BPAP, 5/50 with CPAP
Lesnik et al. [13]	France	Laryngoscope	Retrospective cohort	26 children	<17 yrs	Bilateral vocal cord paralysis	8 treated with CPAP (< 10 yrs)
Nanaware et al. [14]	India	Indian J Pediatr	Retrospective cohort	56 children, 23 with sleep disordered breathing	<18 yrs	52% craniofacial anomaly, 17% NMD or skeletal disease, 1 vocal cord paralysis, 1 achondroplasia, 1 Bardet-Biedl, 1 Albright osteodystrophy	CPAP for 1 patient with bilateral vocal cord paralysis
Leboulanger et al. [15]	France	Pediatrics	Retrospective cohort	7 infants	1-10 m	Pierre Robin Sequence	5 treated with CPAP, 2 with BPAP
Amaddeo et al. [16]	France	Plastic Reconstructive Surgery	Prospective cohort	44 infants	0-2 m	Pierre Robin Sequence	9/44 treated with CPAP
Daniel et al. [17]	Australia	Int J Pediatr Otorhinola	Retrospective cohort	39 infants	> 12 m	Pierre Robin Sequence	18/39 treated with CPAP

		ryngology					
Filip et al. [18]	Norway	Cleft Palate Craniofacial Surgery	Retrospective national cohort 1980-2010	104 children	3.3-33.4 yrs	Pierre Robin Sequence	8 treated with CPAP (duration days to 1 yr), 8 nasopharyngeal tube, 2 oropharyngeal tube, 6 tracheotomy
Kam et al. [19]	Canada	Canadian Respir J	Retrospective cohort 2000-2010	139 patients	9 ± 14 m	139 Pierre Robin Sequence	20 treated with CPAP, 28 nasopharyngeal tube, 45 tong-lip adhesion, 5 mandibular distraction osteogenesis, 19 tracheotomy
Trider et al. [20]	Canada	Int J Pediatr Otorhinolaryngology	Prospective study	51 children	0-14 yrs	CHARGE syndrome	10/51 treated with CPAP
Girbal et al. [21]	Portugal	Rev Port Pneumol	Retrospective study	68 children	1-176 m	5 Pierre Robin Sequence 5 airway malacia 5 Down syndrome 6 Prader Willi syndrome 9 cerebral palsy 10 craniofacial malformation 5 mucopolysaccharidosis 6 metabolic diseases 3 obesity	CPAP (n=52) or NIV (n=16)

Marcus et al. [22]	USA	J Pediatr	Retrospective cohort	94 children	0-19 yrs	25 obesity 23 craniofacial malformation 17 idiopathic OSA 12 Down syndrome 5 cerebral palsy	
Guilleminault et al. [23]	USA	J Pediatr	Retrospective study	74 infants	< 12 m	Syndrome in 38/74: 9 Pierre Robin Sequence, 2 cleft palate, 2 Hunter, 3 achondroplasia, 7 cerebral palsy, 3 epilepsy, 2 hemiplegia, 1 hydrocephalus, 1 NMD	18 discontinued CPAP during follow up
Jarund et al. [24]	Sweden	Scandinavian J Plastic Reconstructive Surgery	Retrospective study	76 children with Apert (27), Crouzon (47), Pfeiffer (2)	23% < 13 yrs	76 children with Apert (27), Crouzon (47), Pfeiffer (2)	7 patients were treated with CPAP after craniofacial surgery
Jarund et al. [25]	Sweden	Scandinavian J Plastic Reconstructive Surgery	Retrospective stduy	13 children with craniofacial malformation, 13 had OSA		1 child had a tracheostomy, 10 of the 12 others accepted CPAP	
Padman et al. [26]	USA	Clinical Pediatr	Retrospective cohort	10 children	3-18 yrs	Obesity and craniofacial malformation	BPAP in 6 obesity 3 craniofacial

							malformation
Massa et al. [27]	UK	Arch Dis Childh	Retrospective cohort 1994-1999	66 children	0.1-19 yrs	24 craniofaciostenosis 6 isolated facial defect 4 osteochondrodysplasia 8 mucopolysaccharidosis, 4 NMD 4 obesity 2 Down syndrome, 2 cerebral palsy 3 airway malacia + other	42 accepted CPAP, 22 refused 3 patients could be weaned after craniofacial surgery
Gonsalez et al. [28]	UK	Childs Nervous System	Retrospective cohort	8 children	2.2-15 yrs	Craniofacial dysostosis	CPAP successful in 5/8
Bannink et al. [29]	The Netherlands	Inter J Oral Maxillofacial Surgery	Retrospective cohort	11 children	4.1-23.2 yrs	Syndromic craniofaciostenosis: 3 Apert, 6 Crouzon, 3 Pfeiffer	3 CPAP or nasopharyngeal tube before mid-face advancement, 3/11 needed CPAP after surgery
Shine et al. [30]	Australia	Arch Otolaryngology Head Neck Surgery	Retrospective cohort	19 children	2-18 yrs	Obesity	10 needed CPAP after AT
Beebe et al.	USA	PlosOne	Prospective	13 obese	14.8 ±	Obesity	

[31]			study	adolescents treated with CPAP for OSA	1.8 yrs	6 were compliant and 7 not	
Puri et al. [32]	USA	J Clin Sleep Med	Retrospective cohort	57 children	1.6 -18 yrs	37 obesity 3 craniofacial malformation	
Konstantinopoulou et al. [33]	USA	Sleep Med	Prospective study	23 children	8-19 yrs	Down syndrome (20 with OSA)	10 children with Down, syndrome were randomized to CPAP
Rosen et al. [34]	USA	Clinical Pediatr	Retrospective study	29 infants with Down syndrome, 16 had OSA	< 2 yrs	Down syndrome	6 treated with CPAP Spontaneous improvement in 3 infants after 5 to 10 m
Shete et al. [35]	USA	Int J Pediatr Otorhinolaryngology	Retrospective study	AT in 11 children with Down syndrome + 11 controls	mean age 8.5 yrs	Down syndrome	6 required CPAP or BPAP after AT
Esbensen et al. [36]	USA	J Dev Behav Pediatr	Retrospective study	954 children 455 (47.7%) had a PSG	5-25 yrs	Down syndrome 258 (27%) had OSA:	Patients with OSA: 18.6% treated with CPAP 82 (8.6%) had OSA + behavior disorder: 19.5% treated with CPAP
Dudoignon	France	Am J Med	Retrospective	57 T21	6.2 ± 5.9	Down syndrome	15/57 treated with CPAP

et al. [37]		Genetics Part A	study		yrs		for OSA 4/57 treated with NIV for hypoventilation
Amaddeo et al. [38]	France	Pediatr Pulmonol	Retrospective study	31 children started on CPAP in an out-patient setting	0.8 m-17.5 yrs	7 Down syndrome, 3 achondroplasia, 3 obesity and other	4 (3 Down syndrome) not compliant 3 patients weaned from CPAP
Sudarsan et al. [39]	India	Int J Pediatr Otorhinolaryngology	Prospective study, efficacy AT in mucopolysaccharidosis and Down syndrome	73 children	6-12 yrs	Mucopolysaccharidosis Down syndrome	17 mucopolysaccharidosis and 29 Down syndrome treated with CPAP
Tirosh et al. [40]	Israel	Acta Pediatr	Description of 4 patients	4 children with neurodisability	6 - 16 yrs	1 obesity + mental retardation, 1 attention-deficit/hyperactivity disorder, 1 epilepsy, 1 fragile X syndrome	CPAP possible in children with neurodisability
Al-Iede et al. [41]	Australia	Sleep Med	Retrospective study	148 children treated with CPAP for non-OSA	0.1-16.8 yrs (64% < 1yr)	47 chronic lung disease 37 congenital heart disease, 41 laryngomalacia, 16 tracheobronchomalacia,	

						5 congenital diaphragmatic hernia	
Kirk et al. [42]	Canada	Pediatr Pulmonol	Retrospective study	73 children	0-18 yrs	Myelomeningocele	30 OSA: CPAP success in 18/21 25 CSA: NIV required in 7
Khirani et al [43]	France	Crit Care	Prospective study	12 infants	2-22 m	5 BPD 4 laryngomalacia 1 OSA, 1 Down syndrome, 1 Pierre Robin Sequence	
Waters et al. [44]	Australia	AJRCCM	Retrospective study	80 children	5.7 ± 0.5 yrs	53% of children had a congenital syndrome or malformation	
Clift et al. [45]	UK	J Sleep Research	Retrospective study	17 children and young adults	?	Prader Willi syndrome	7/17 obese patients were treated with CPAP or BPAP (2 did not tolerate)
Pavone et al. [46]	Italy and France	Pediatr Pulmonol	Retrospective study	70 children (< 20 yrs) and 18 adults		Prader Willi syndrome	16/88 treated with CPAP or BPAP (> older patients)
Facchina et al. [47]	France	Am J Med Genetics Part A	Retrospective study	16 children	10.5 ± 4.2 yrs	Mucopolysaccharidosis type IVA (Morquio)	4/16 treated with CPAP (OSA) 2./16 treated with NIV (hypoventilation)
Tabone et	France	Am J Med	Retrospective	7 children	0.3-17.4	7 mucolipidosis	5 CPAP

al. [48]		Genetics	study		yrs	5 MLII, 1 type II-III, 1 type III: OSA in 6/7	1 NIV (hypoventilation)
Leotard et al. [49]	France	Ann Phys Rehabil Med	Retrospective study	15/188 patients with osteogenesis imperfecta had a PSG		12/15 patients with osteogenesis imperfecta had sleep disordered breathing:	2 patients treated with CPAP
Khirani et al. [50]	France	Am J Med Genetics Part A	Retrospective study	10 children	3.3-14.1 yrs	10 pycnodysostosis	9/10 treated with CPAP Weaning in 3 patients, 2 after surgery and 1 spontaneous
Rosen et al. [51]	USA	Supportive Care in Cancer	Retrospective study	70 children with cancer, PSG in 53, 9/20 severe OSA	6-21 yrs	Neoplasms of the CNS	9 treated with CPAP (6 success) and 3 with brainstem tumor treated with BPAP
Bunn et al. [52]	UK	Pediatr Cardiology	Retrospective study	4 children	3-34 m	Congenital cardiopathy and PHT	NIV for 13 – 19 m, correction of PHT with NIV
Domany et al. [53]	USA	J Clin Sleep Med	Retrospective study	65 children	< 18 yrs	Children with Ehlers Danlos referred to a sleep lab 17 OSA, 2 treated with CPAP (1 with mild OSA + somnolence and 1 with	No detailed information on CPAP

						moderate who refused AT)	

Abbreviations: m: months, yrs: years, OSA: obstructive sleep apnea, BPAP: bilevel positive airway pressure, NIV: noninvasive ventilation, Poeso: oesophageal pressure, Pgas: gastric pressure, NMD: neuromuscular disease, BPD: bronchopulmonary dysplasia, PSG: polysomnography, CNS: central nervous system, PHT: pulmonary hypertension, AT: adenotonsillectomy

References

1. Afsharpaiman S, Sillence DO, Sheikhvatan M, *et al.* Respiratory events and obstructive sleep apnea in children with achondroplasia: investigation and treatment outcomes. *Sleep Breath* 2011; 15: 755-761.
2. Mogayzel PJJ, Carroll JL, Loughlin GM, *et al.* Sleep-disordered breathing in children with achondroplasia. *J Pediatr* 1998; 132: 667-671.
3. Schlüter B, De Sousa G, Trowitzsch E, *et al.* Diagnostics and management of sleep-related respiratory disturbances in children with skeletal dysplasia caused by FGFR3 mutations (achondroplasia and hypochondroplasia). *Georgian Med News* 2011; 63-72.
4. Waters KA, Everett F, Sillence DO, *et al.* Treatment of obstructive sleep apnea in achondroplasia: evaluation of sleep, breathing, and somatosensory-evoked potentials. *Am J Med Genet* 1995; 59: 460-466.
5. Julliand S, Boule M, Baujat G, *et al.* Lung function, diagnosis, and treatment of sleep-disordered breathing in children with achondroplasia. *Am J Med Genet A* 2012; 158A: 1987-1993.
6. Tenconi R, Khirani S, Amaddeo A, *et al.* Sleep-disordered breathing and its management in children with achondroplasia. *Am J Med Genet A* 2017; 173: 868-878.
7. Fauroux B, Pigeot J, Polkey MI, *et al.* Chronic stridor caused by laryngomalacia in children: work of breathing and effects of noninvasive ventilatory assistance. *Am J Respir Crit Care Med* 2001; 164: 1874-1878.
8. Zwacka G, Scholle S, Kemper G, *et al.* Nasal CPAP therapy for infants with congenital stridor. *Sleep Breath* 1997; 2: 85-97.
9. Essouri S, Nicot F, Clement A, *et al.* Noninvasive positive pressure ventilation in infants with upper airway obstruction: comparison of continuous and bilevel positive pressure. *Intensive Care Med* 2005; 31: 574-580.
10. Kawaguchi AL, Donahoe PK, Ryan DP. Management and long-term follow-up of patients with types III and IV laryngotracheoesophageal clefts. *J Pediatr Surg* 2005; 40: 158-165.
11. Pellen G, Pandit C, Castro C, *et al.* Use of non-invasive ventilation in children with congenital tracheal stenosis. *Int J Pediatr Otorhinolaryngol* 2019; 127: 109672.

12. Shatz A, Goldberg S, Picard E, *et al.* Pharyngeal wall collapse and multiple synchronous airway lesions. . *Ann Otol Rhinol Laryngol* 2004; 113: 483-487.
13. Lesnik M, Thierry B, Glynn F, *et al.* Idiopathic bilateral vocal cord paralysis in infants: Case series and literature review. *Laryngoscope* 2015; 125: 1724-1728.
14. Nanaware SKV, Gothi D, Joshi JM. Sleep apnea. *Indian J Pediatr* 2006; 73: 597-601.
15. Leboulanger N, Picard A, Soupre V, *et al.* Physiologic and clinical benefits of noninvasive ventilation in infants with Pierre Robin sequence. *Pediatrics* 2010; 126: e1056-1063.
16. Amaddeo A, Abadie V, Chalouhi C, *et al.* Continuous positive airway pressure for upper airway obstruction in infants with Pierre Robin Sequence. *Plast Reconstruct Surg* 2016; 137: 609-612.
17. Daniel M, Bailey S, Walker K, *et al.* Airway, feeding and growth in infants with Robin sequence and sleep apnoea. *Int J Pediatr Otorhinolaryngol* 2013; 77: 499-503.
18. Filip C, Feragen KB, Lemvik JS, *et al.* Multidisciplinary aspects of 104 patients with Pierre Robin Sequence. *Cleft Palate Craniofac J* 2015; 52: 732-742.
19. Kam K, McKay M, MacLean J, *et al.* Surgical versus nonsurgical interventions to relieve upper airway obstruction in children with Pierre Robin sequence. *Can Respir J* 2015; 22: 171-175.
20. Trider CL, Corsten G, Morrison D, *et al.* Understanding obstructive sleep apnea in children with CHARGE syndrome. *Int J Pediatr Otorhinolaryngol* 2012; 76: 947-953.
21. Girbal IC, Goncalves C, Nunes T, *et al.* Non-invasive ventilation in complex obstructive sleep apnea--a 15-year experience of a pediatric tertiary center. *Rev Port Pneumol* 2014; 20: 146-151.
22. Marcus CL, Ward SL, Mallory GB, *et al.* Use of nasal continuous positive airway pressure as treatment of childhood obstructive sleep apnea. *J Pediatr* 1995; 127: 88-94.
23. Guilleminault C, Pelayo R, Clerk A, *et al.* Home nasal continuous positive airway pressure in infants with sleep-disordered breathing. *J Pediatr* 1995; 127: 905-912.
24. Jarund M, Lauritzen C. Craniofacial dysostosis: Airway obstruction and craniofacial surgery. *Scand J Plast Reconstr Surg Hand Surg* 1996; 30: 275-279.
25. Jarund M, Dellborg C, Carlson J, *et al.* Treatment of sleep apnoea with continuous positive airway pressure in children with craniofacial malformations. *Scand J Plast Reconstr Surg Hand Surg* 1999; 33: 67-71.
26. Padman R, Hyde C, Foster P, *et al.* The pediatric use of bilevel positive airway pressure therapy for obstructive sleep apnea syndrome: a retrospective review with analysis of respiratory parameters. *Clin Pediatr* 2002; 41: 163-169.
27. Massa F, Gonsalez S, Laverty A, *et al.* The use of nasal continuous positive airway pressure to treat obstructive sleep apnoea. *Arch Dis Child* 2002; 87: 438-443.

28. Gonsalez S, Thompson D, Hayward R, *et al.* Treatment of obstructive sleep apnoea using nasal CPAP in children with craniofacial dysostoses. *Childs Nerv Syst* 1996; 12: 713-719.
29. Bannink N, Nout E, Wolvius EB, *et al.* Obstructive sleep apnea in children with syndromic craniosynostosis: long-term respiratory outcome of midface advancement. *Int J Oral Maxillofac Surg* 2010; 39: 115-121.
30. Shine NP, Lannigan FJ, Coates HL, *et al.* Adenotonsillectomy for obstructive sleep apnea in obese children: effects on respiratory parameters and clinical outcome. *Arch Otolaryngol Head Neck Surg* 2006; 132: 1123-1127.
31. Beebe DW, Byars KC. Adolescents with obstructive sleep apnea adhere poorly to positive airway pressure (PAP), but PAP users show improved attention and school performance. *PLoS One* 2011; 6: e16924.
32. Puri P, Ross KR, Mehra R, *et al.* Pediatric positive airway pressure adherence in obstructive sleep apnea enhanced by family member positive airway pressure usage. *J Clin Sleep Med* 2016; 12: 959-963.
33. Konstantinopoulou S, Tapia IE, Kim JY, *et al.* Relationship between obstructive sleep apnea cardiac complications and sleepiness in children with Down syndrome. *Sleep Med* 2016; 17: 18-24.
34. Rosen D. Some infants with Down syndrome spontaneously outgrow their obstructive sleep apnea. *Clin Pediatr* 2010; 49: 1068-1071.
35. Shete MM, Stocks RMS, Sebelik ME, *et al.* Effects of adeno-tonsillectomy on polysomnography patterns in Down syndrome children with obstructive sleep apnea: a comparative study with children without Down syndrome. *Int J Pediatr Otorhinolaryngol* 2010; 74: 241-244.
36. Esbensen AJ, Beebe DW, Byars KC, *et al.* Use of sleep evaluations and treatments in children with Down syndrome. *J Dev Behav Pediatr* 2016; 37: 629-636.
37. Dudoignon B, Amaddeo A, Frapin A, *et al.* Obstructive sleep apnea in Down syndrome: Benefits of surgery and noninvasive respiratory support. *Am J Med Genet A* 2017; 173: 2074-2080.
38. Amaddeo A, Frapin A, Touil S, *et al.* Outpatient initiation of long-term continuous positive airway pressure in children. *Pediatr Pulmonol* 2018; 53: 1422-1428.
39. Sudarsan SS, Paramasivan VK, Arumugam SV, *et al.* Comparison of treatment modalities in syndromic children with obstructive sleep apnea--a randomized cohort study. *Int J Pediatr Otorhinolaryngol* 2014; 78: 1526-1533.
40. Tirosh E, Tal Y, Jaffe M. CPAP treatment of obstructive sleep apnoea and neurodevelopmental deficits. *Acta Paediatr* 1995; 84: 791-794.
41. Al-Iede M, Kumaran R, Waters K. Home continuous positive airway pressure for cardiopulmonary indications in infants and children. *Sleep Med* 2018; 48: 86-92.
42. Kirk VG, Morielli A, Gozal D, *et al.* Treatment of sleep-disordered breathing in children with myelomeningocele. *Pediatr Pulmonol* 2000; 30: 445-452.
43. Khirani S, Ramirez A, Aloui S, *et al.* Continuous positive airway pressure titration in infants with severe upper airway obstruction or bronchopulmonary dysplasia. *Crit Care* 2013; 17: R167.
44. Waters KA, Everett FM, Bruderer JW, *et al.* Obstructive sleep apnea: the use of nasal CPAP in 80 children. *Am J Respir Crit Care Med* 1995; 152: 780-785.

45. Clift S, Dahlitz M, Parkes JD. Sleep apnoea in the Prader-Willi syndrome. *J Sleep Res* 1994; 3: 121-126.
46. Pavone M, Caldarelli V, Khirani S, *et al.* Sleep disordered breathing in patients with Prader-Willi syndrome: A multicenter study. *Pediatr Pulmonol* 2015; 50: 1354-1359.
47. Facchina G, Amaddeo A, Baujat G, *et al.* A retrospective study on sleep-disordered breathing in Morquio-A syndrome. *Am J Med Genet A* 2018; 176: 2595-2603.
48. Tabone L, Caillaud C, Amaddeo A, *et al.* Sleep-disordered breathing in children with mucolipidosis. *Am J Med Genet A* 2019; 179: 1196-1204.
49. Léotard A, Taytard J, Aouate M, *et al.* Diagnosis, follow-up and management of sleep-disordered breathing in children with osteogenesis imperfecta. *Ann Phys Rehabil Med* 2018; 61: 135-139.
50. Khirani S, Amaddeo A, Baujat G, *et al.* Sleep-disordered breathing in children with pycnodysostosis. *Am J Med Genet A* 2020; 182: 122-129.
51. Rosen G, Brand SR. Sleep in children with cancer: case review of 70 children evaluated in a comprehensive pediatric sleep center. *Support Care Cancer* 2011; 19: 985-994.
52. Bunn HJ, Roberts P, Thomson AH. Noninvasive ventilation for the management of pulmonary hypertension associated with congenital heart disease in children. *Pediatr Cardiol* 2004; 25: 357-359.
53. Domany KA, Hantragool S, Smith DF, *et al.* Sleep disorders and their management in children with Ehlers-Danlos syndrome referred to sleep clinics. *J Clin Sleep Med* 2018; 14: 623-629.