

Table 37: Excluded clinical studies for interventions for dystonia

Excluded studies - A.3 Which treatments (pharmacological treatment (levodopa, anticholinergic drugs, and botulinum toxin injections), neurosurgical procedure (deep brain stimulation, ITB) are most effective for managing dystonia in adults with cerebral palsy where dystonia is the predominant abnormality of tone?	
Study	Reason for Exclusion
Beecham, E., Candy, B., Howard, R., McCulloch, R., Laddie, J., Rees, H., Vickerstaff, V., Bluebond-Langner, M., Jones, L., Pharmacological interventions for pain in children and adolescents with life-limiting conditions, Cochrane Database of Systematic Reviews, 3, CD010750, 2015	Systematic review but in children- includes Bonouvrie 2011 and Hoving 2007 intrathecal baclofen trials
Berweck, S., BP-DBS for dystonia-choreoathetosis cerebral palsy, Lancet Neurology, 8, 692-693, 2009	Editorial on Vidailhet 2009
Bonouvrie, L. A., Becher, J. G., Vles, J. S. H., Boeschoten, K., Soudant, D., de Groot, V., van Ouwerkerk, W. J. R., Strijers, R. L. M., Foncke, E., Geytenbeek, J., van de Ven, P. M., Teernstra, O., Vermeulen, R. J., Intrathecal baclofen treatment in dystonic cerebral palsy: A randomized clinical trial: The IDYS trial, BMC Pediatrics, 13, 2013	Trial protocol for IDYS RCT
Bonouvrie, L., Becher, J., Soudant, D., Buizer, A., Van Ouwerkerk, W., Vles, G., Vermeulen, R. J., The effect of intrathecal baclofen treatment on activities of daily life in children and young adults with cerebral palsy and progressive neurological disorders, European Journal of Paediatric Neurology, 20, 538-544, 2016	No measurements before ITB - patients/carers asked to recall the situation before ITB pump
Boyd, Rn, Dobson, F, Parrott, J, Love, S, Oates, J, Larson, A, Burchall, G, Chondros, P, Carlin, J, Nattrass, G, Graham, Hk, The effect of botulinum toxin type A and a variable hip abduction orthosis on gross motor function: a randomized controlled trial, European Journal of Neurology, 8 Suppl 5, 109-19, 2001	Not dystonia, age 1-4 yrs
Butler, C., Campbell, S., Evidence of the effects of intrathecal baclofen for spastic and dystonic cerebral palsy, Developmental Medicine & Child Neurology, 42, 634-645, 2000	Systematic review, outdated - includes Albright 1996
Cif, A. L., Biolsi, B., Robles, S. G., El Fertit, H., Tancu, C., Vasquez, X., Coubes, P., Internal globus pallidus stimulation in the treatment of dystonic and dyskinesic syndromes associated with cerebral palsy, European Journal of Neurology, 13, 74-74, 2006	Abstract only - reports mean improvement in Burke Fahn Marsdenâ™s Dystonia Rating Scale

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Study	Reason for Exclusion
Cif, L., Deep brain stimulation in dystonic cerebral palsy: for whom and for what?, <i>European Journal of Neurology</i> , 22, 423-425, 2015	Editorial on Romita study
Cif, L., Martinez, V. G., Sanrey, E., Nerrant, E., Ros, M., Cyprien, F., Seng, E. C., Roujeau, T., Coubes, P., Axial prominent, delayed onset dystonia in cerebral palsy: Highlights on a distinct phenotype with favorable outcome following deep brain stimulation, <i>Movement Disorders</i> , 31, S561-S561, 2016	Abstract only, case series (N=5)
Coelho, M., Cattoni, B., Lobo, P. P., Carvalho, H., Guedes, L. C., Sousa, P. R., Grunho, M., Albuquerque, L., Pereira, J. M., Reimao, S., Morgado, C., Ferreira, J. J., Rosa, M. M., Ferreira, A. G., Deep brain stimulation for the treatment of primary dystonias and dyskinetic cerebral palsy, <i>European Journal of Neurology</i> , 19, 678-678, 2012	Abstract only, case series (N=5)
Eltahawy, H. A., Saint-Cyr, J., Giladi, N., Lang, A. E., Lozano, A. M., Primary dystonia is more responsive than secondary dystonia to pallidal interventions: outcome after pallidotomy or pallidal deep brain stimulation, <i>Neurosurgery</i> , 54, 613-19; discussion 619-21, 2004	Not cerebral palsy
Gaasterland, C. M. W., Jansen-van der Weide, M. C., Weinreich, S. S., van der Lee, J. H., A systematic review to investigate the measurement properties of goal attainment scaling, towards use in drug trials, <i>BMC Medical Research Methodology</i> <i>BMC Med Res Methodol</i> , 16, 2016	Systematic review of trials using goal attainment scales as outcomes
Katsakiori, P. F., Kefalopoulou, Z., Markaki, E., Paschali, A., Ellul, J., Kagadis, G. C., Chroni, E., Constantoyannis, C., Deep brain stimulation for secondary dystonia: results in 8 patients, <i>Acta Neurochirurgica</i> , 151, 473-478, 2009	N=2 patients with cerebral palsy
Kerr, C., McDowell, B., Cosgrove, A., Walsh, D., Bradbury, I., McDonough, S., Electrical stimulation in cerebral palsy: a randomized controlled trial, <i>Developmental Medicine and Child Neurology</i> , 48, 870-6, 2006	neuromuscular electrical stimulation, age < 16 years
Kerr, C., McDowell, B., McDonough, S., Electrical stimulation in cerebral palsy: A review of effects on strength and motor function, <i>Developmental Medicine and Child Neurology</i> , 46, 205-213, 2004	Systematic review of neuromuscular electrical stimulation
Kim, J. P., Chang, W. S., Chang, J. W., Treatment of secondary dystonia with a combined stereotactic procedure: long-term surgical outcomes, <i>Acta Neurochirurgica</i> , 153, 2319-27; discussion 2328, 2011	Combines DBS with unilateral thalamotomy
Koy, A., Hellmich, M., Pauls, A. M., Marks, W. A., Lin, J. P., Fricke, O., Timmermann, L., The effect of deep brain stimulation on cerebral palsy: A meta-analysis, <i>Movement Disorders</i> , 27, S318-S318, 2012	Systematic review - does not report by age subgroups. Checked for relevant studies.
Lannin, N. A., Novak, I., Cusick, A., A systematic review of upper extremity casting for children and adults with central	Systematic review= no studies of adults with CP

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Study	Reason for Exclusion
nervous system motor disorders, Clinical Rehabilitation, 21, 963-76, 2007	
Lettieri, C., Rinaldo, S., Devigili, G., Pisa, F., Mucchiut, M., Belgrado, E., Mondani, M., D'Auria, S., Ius, T., Skrap, M., Eleopra, R., Clinical outcome of deep brain stimulation for dystonia: Constant-current or constant-voltage stimulation? A non-randomized study, European Journal of Neurology, 22, 919-926, 2015	Mixed population of primary and other dystonia - cerebral palsy not reported.
Marks, W., Honeycutt, J., Acosta, F., Bailey, L., Reed, M., Pomykal, A., Mercer, M., Pallidal Stimulation Improves Dystonia in Cerebral Palsy, Neurology, 76, A330-A331, 2011	Abstract only - See Marks 2011 for full text
McGinley, J., Dobson, F., Morgan, P., A systematic review of the effect of interventions on gait in adults with cerebral palsy, Developmental Medicine and Child Neurology, 54, 45-46, 2012	Systematic review - interventions not in protocol (more relevant for topic D2)
Merrill, D.R., Review of electrical stimulation in cerebral palsy and recommendations for future directions, Developmental Medicine and Child Neurology, 51, 154-165, 2009	Review of neuromuscular electrical stimulation
Mueller, J., Skogseid, I. M., Benecke, R., Kupsch, A., Trottenberg, T., Poewe, W., Schneider, G. H., Eisner, W., Wolters, A., Muller, J. U., Deuschl, G., Pinsker, M. O., Roeste, G. K., Vollmer-Haase, J., Brentrup, A., Krause, M., Tronnier, V., Schnitzler, A., Voges, J., Nikkhah, G., Vesper, J., Naumann, M., Volkmann, J., Pallidal deep brain stimulation improves quality of life in segmental and generalized dystonia: Results from a prospective, randomized sham-controlled trial, Movement Disorders, 23, 131-134, 2008	Primary dystonia
Naumann, M., Jankovic, J., Safety of botulinum toxin type A: a systematic review and meta-analysis, Current Medical Research and Opinion, 20, 981-990, 2004	Systematic review - some studies were in cerebral palsy but the age of participants and indication for botox not reported.
Pin, T. W., McCartney, L., Lewis, J., Waugh, M. C., Use of intrathecal baclofen therapy in ambulant children and adolescents with spasticity and dystonia of cerebral origin: a systematic review, Developmental Medicine & Child Neurology, 53, 885-95, 2011	Systematic review- found no studies on the use of ITB in children/adolescents with dystonia of cerebral origin
Romito, L. M., Zorzi, G., Ciceri, M. L., Marras, C. E., Franzini, A., Nardocci, N., Albanese, A., Long-term follow-up of GPi deep brain stimulation in generalized dystonia: Primary dystonia compared to cerebral palsy, Movement Disorders, 28, S434-S434, 2013	Abstract only - see Romito 2015
Schjerling, L., Hjermand, L. E., Jespersen, B., Madsen, F. F., Brennum, J., Jensen, S. R., Lokkegaard, A., Karlsborg, M., A randomized double-blind crossover trial comparing subthalamic and pallidal deep brain stimulation for dystonia: Clinical article, Journal of Neurosurgery, 119, 1537-1545, 2013	Primary dystonia or secondary to medication.
Sokal, P., Rudas, M., Harat, M., Szyberg, L., Zielinski, P., Deep anterior cerebellar stimulation reduces symptoms of	Predominantly spastic CP with secondary dystonia – examines

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Study	Reason for Exclusion
secondary dystonia in patients with cerebral palsy treated due to spasticity, <i>Clinical Neurology and Neurosurgery</i> , 135, 62-68, 2015	deep anterior cerebellar stimulation.
Sommerfelt, K, Markestad, T, Berg, K, Saetesdal, I, Therapeutic electrical stimulation in cerebral palsy: a randomized, controlled, crossover trial, <i>Developmental Medicine and Child Neurology</i> , 43, 609-13, 2001	Neuromuscular stimulation - age <12 years
Ubhi, T, Bhakta, Bb, Ives, Hl, Allgar, V, Roussounis, Sh, Randomised double blind placebo controlled trial of the effect of botulinum toxin on walking in cerebral palsy, <i>Archives of Disease in Childhood</i> , 83, 481-7, 2000	Not predominantly dystonic cerebral palsy. Age <16 years
Volkman, J., Wolters, A., Kupsch, A., Muller, J., Kuhn, A. A., Schneider, G. H., Poewe, W., Hering, S., Eisner, W., Muller, J. U., Deuschl, G., Pinsker, M. O., Skogseid, I. M., Roeste, G. K., Krause, M., Tronnier, V., Schnitzler, A., Voges, J., Nikkhah, G., Vesper, J., Classen, J., Naumann, M., Benecke, R., Pallidal deep brain stimulation in patients with primary generalised or segmental dystonia: 5-year follow-up of a randomised trial, <i>The Lancet Neurology</i> , 11, 1029-1038, 2012	Primary dystonia
Walker, R. H., Danisi, F. O., Swope, D. M., Goodman, R. R., Germano, I. M., Brin, M. F., Intrathecal baclofen for dystonia: Benefits and complications during six years of experience, <i>Movement Disorders</i> , 15, 1242-1247, 2000	1/14 included had CP
Wloch, A., Abdallat, M., Saryyeva, A., Blahak, C., Wolf, J., Schrader, C., Runge, J., Krauss, J. K., Complications of deep brain stimulation for secondary dystonia in the early postoperative period (30-day morbidity): An experience in 49 patients, <i>Journal of Neural Transmission</i> , 123 (12), 1525, 2016	Abstract only. 17/49 had CP - their results are not reported separately
Wong, C, Pedersen, Sa, Kristensen, Bb, Gosvig, K, Sonne-Holm, S, The Effect of Botulinum Toxin A Injections in the Spine Muscles for Cerebral Palsy Scoliosis, Examined in a Prospective, Randomized Triple-blinded Study, <i>Spine</i> , 40, E1205-11, 2017	Not dystonia, age <=18 years