Pelvic-Spinal Analysis and the Impact of Onabotulinum toxin A Injections on Spinal Balance in one Child With Cerebral Palsy

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Abstract

Background: In children with cerebral palsy, primary (eg, abnormal muscle tone and weakness) and secondary impairments (eg, contractures) can modify pelvic-spinal alignment. The main aim of this article was to establish a new approach to pelvic-spinal analysis in children with cerebral palsy, taking into account the whole pelvis-spine complex, illustrated by a case study. **Methods:** This is a case study of an ambulatory child with cerebral palsy (spastic diplegia) who underwent analysis of the pelvic-spine complex from X-ray images taken in standing position from C2 to the proximal femur. Pelvic shape was characterized by the pelvic incidence angle, which is the sum of sacral slope and pelvic tilt, before and after the treatment by regular onabotulinumtoxinA injections into the hip flexors, and the use of soft lumbar brace over 5 years. **Results:** The sagittal balance of the spine was improved following the treatment, with a reduction in lumbar lordosis and sacral slope. The reduction in lumbar hyperextension likely reduced the risk of spondylolysis, low back pain, and degenerative spondylolisthesis in adulthood. **Conclusion:** A biomechanical approach to the evaluation of the pelvic-spinal complex offers new perspectives to increase the understanding of spinal balance in children with cerebral palsy, providing more options for treatment, such as onabotulinumtoxinA.

Keywords

cerebral palsy, botulinum toxin, children, assessment, pelvic-spinal balance

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Botulinum toxin has been used for more than 15 years to reduce muscle tone in children with cerebral palsy and is considered as the standard treatment for spasticity.^{1,2} The decrease in muscle tone increases range of motion and strength of antagonist muscles, improves the gait pattern, and reduces pain.³ In children with cerebral palsy, the goal of treatment is often the prevention of complications. The treatment of primary impairments (such as abnormal muscle tone) can prevent the occurrence of secondary impairments (such as muscle and tendon contractures and bony deformities) and tertiary problems (such as abnormal posture), thereby reducing the long-term impact of the pathology.⁴

Bony deformities, including spinal deformities, develop during childhood and adolescence and can cause pain and reduced mobility in adulthood. However, deformities may be prevented by injections of botulinum toxin during childhood.⁵ During growth, the development and alignment of the spine may be affected by primary and secondary impairments, including bony anomalies in the lower limbs. Children with cerebral palsy often have an abnormal spinal alignment compared to their healthy peers. In the lumbar spine, 2 main abnormal sagittal posture types can be found, depending on the

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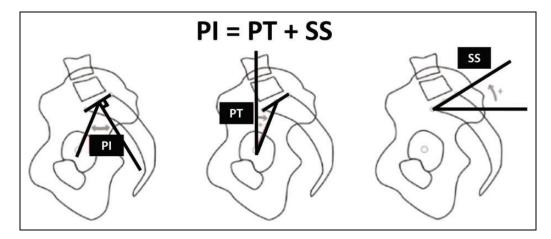


Figure 1. Pelvic parameters: 3 angles (Duval-Beaupère et al, 1992⁶). Pl indicates pelvic incidence; PT, pelvic tilt; SS, sacral slope.

pattern of neuromotor impairment—hyperlordosis of the lumbar spine associated with an exaggerated anterior pelvic tilt that is difficult to correct by conservative or surgical methods (as described by Song et al⁷) and hypolordosis of the lumbar spine associated with posterior pelvic tilt and contracture of the hamstring muscles, which also causes an increase in the popliteal angle (as described by Mac Carthy et al⁸).

Botulinum toxin injections in the lower limb muscles could change the global pelvis-spine balance; however, there are currently no published guidelines for this treatment since few studies have specifically considered the consequences of cerebral palsy on pelvis-spine balance.⁹ Equally, few objective measures exist to evaluate the baseline state and to determine changes following treatment.

The main aim of this article was therefore to establish a new approach to spinal analysis in children with cerebral palsy, taking into account the whole pelvis-spine complex, illustrated by a case study.

Method

A case study of a female child with spastic diplegia named L., based on the assessments and treatments she underwent between the ages of 5 and 13 years. At 5 years of age, her functional level was rated as 1 on the Gross Motor Function Classification System, and she walked with a true equinus gait pattern (type I), with persistence of ankle plantar flexion in standing and moderate genu recurvatum. Her pelvis was anteriorly tilted with a compensatory hyperlordosis. The ground reaction force was anterior to the knee and hip, with anterior pelvic tilt and lumbar hyperlordosis to compensate for the posterior instability.

Treatment From Age 5 to 8 Years—OnabotulinumtoxinA Injections of Gastrocnemius and Soleus

Significant spasticity was found only in the gastrocnemius and soleus muscles, with a full range of motion and good selective motor control of the antagonist muscles. It was thus hypothesized that the exaggerated anterior tilt of the pelvis was a compensation for the equinus gait. OnabotulinumtoxinA was administered in the 3 triceps surae heads (dilution 50 U/mL). A total dose of 210 U was injected—40 U in the medial and lateral gastrocnemius of each limb (80 U per limb, total 160 U) and 25 U in the left and right soleus (25 U per limb, total 50 U).

Following the treatment, the gait pattern improved, including heel contact on the ground during standing. Injections were thus continued at 6-month intervals in the same muscles, with the same total dose but smaller doses in the soleus. This treatment was combined with a strengthening program, particularly of the gluteus maximus muscle. After 3 years, there was no change in anterior pelvic tilt, although the position of the force vector in standing had improved.

Evaluation at 8 Years

Pelvic-spinal analysis. Since reduction of the equinus had not improved the pelvic position, it was decided to investigate the pelvis and spine more closely using radiographic measures. The parameters measured have been described in detail by Roussouly et al¹⁰ and Berthonnaud et al.¹¹

The pelvic parameters define the geometry¹⁰ and spatial position of the pelvis. Three angles that are geometrically related were analyzed—pelvic incidence, pelvic tilt, and sacral slope (Figure 1). Lumbar lordosis was analyzed for the spine.

Pelvic incidence does not change with the position of the patient. It is considered constant after 9 to 11 years of age.¹² Normal values range from 35° to 85° , with a mean value of $50^{\circ} \pm 10^{\circ}$. Pelvic tilt angle describes the amount of rotation of the pelvis around the femoral heads.

Sacral slope is complementary to pelvic tilt. The sum of sacral slope and pelvic tilt is equal to pelvic incidence. Although pelvic incidence is constant, pelvic tilt and sacral slope are variable positional parameters, so when pelvic tilt increases, sacral slope decreases and vice versa.

Lumbar lordosis was defined as the segment between S1 distally and the inflection point where the spine transitions

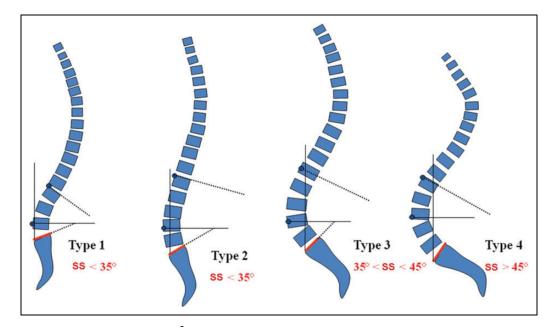


Figure 2. Lumbopelvic morphotype classification.⁹

from lordosis to kyphosis proximally. Its apex is the most anterior point of the lordosis, with the spine in an upright position. Thus, the lumbar lordosis could be considered as long (including thoracic vertebra) or short, as well as deep or shallow. The normal angle of lumbar lordosis is $48.0^{\circ} \pm 11.7^{\circ.8}$

Global analysis of spinal balance. A vertical line from C7 should pass through the sacral end plate,¹³ usually the posterior aspect, and then descend behind the femoral heads. This line is parallel to the line of gravity and is highly correlated with it, thus enabling the evaluation of global spinal balance in clinical practice.

Spinal alignment was classified according to the 4 types described by Roussouly et al^{10} (Figure 2):

- Type 1: (sacral slope <35°), the apex is very low, close to L5; the lumbar lordosis is short and the thoracic kyphosis is long, extending down to the thoraco-lumbar area.
- Type 2: (sacral slope <35°), the distal arc is flat, close to a straight line; the lumbar lordosis is shallow and long. The vertebral column is relatively straight, without curves.
- Type 3: (35° < sacral slope < 45°), the lumbar lordosis is normal, well balanced, and the apex is located in the center of L4. The curves of the vertebral column are regular. This is the most common pattern in asymptomatic participants.
- Type 4: (sacral slope >45°), the apex is very high, at the upper limit of L4 or even higher, the lumbar lordosis is very deep with an inflection point far above T12. The lumbar spine is hyperlordotic, with an increased thoracic kyphosis to compensate.

Results

Pelvic-Spinal Analysis Before OnabotulinumtoxinA Injection in the Psoas—Age 8

The angles of pelvic incidence, pelvic tilt, sacral slope, and lumbar lordosis are shown in Figure 3A. The angle of pelvic incidence was normal, but the lumbar lordosis angle was too high in relation to the pelvic incidence, and the pelvic tilt was negative, inducing a high sacral slope value and causing anterior imbalance, that is, forward projection of the trunk. The vertical line from C7 to the sacral end plate passed in front of the femoral heads.

Spinal alignment type according to Roussouly's classification resembled a type IV (sacral slope >45°), but was not a real type IV, because of the normal pelvic incidence; there was thus a mismatch between the high lumbar lordosis and sacral slope values and the normal pelvic incidence.

The clinical evaluation showed that there was at this stage no spasticity but only some viscoelastic disorders in the gastrocnemius/soleus muscles, and spasticity of the psoas muscle had increased to 2 on the modified Ashworth scale. Since the psoas is a strong muscle that can anteriorly tilt the pelvis, spasticity of this muscle could play a role in the abnormal posture of the pelvis in L.

Treatment

Subsequent onabotulinumtoxinA injections were carried out in the psoas muscles at 6-month intervals. The proximal part of the psoas belly was injected using a posterior approach under both electromyography and ultrasound guidance. A dose of 75 U was injected in each psoas, diluted to 50 U/mL (total dose 150 U). The gastrocnemii and soleus were not injected. In

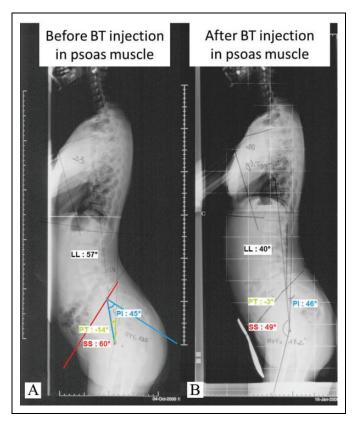


Figure 3. Case study: L.—Sagittal radiographs of the whole spine, before (A) and after BT injection (B) in psoas muscle combined with a soft lumbar brace. BT, botulinum toxin; LL, lumbar lordosis; PI, pelvic incidence; PT, pelvic tilt; SS, sacral slope.

addition, L. wore a soft lumbar brace 5 days per week to reduce the lordosis.

Pelvic-Spinal Analysis After OnabotulinumtoxinA Injection of the Psoas—Age 13

A new pelvic-spinal complex analysis was conducted when L. was 13 years of age. The angles of pelvic incidence, pelvic tilt, sacral slope, and lumbar lordosis are shown in Figure 3B. The value of the pelvic incidence was similar (as expected since this is a constant parameter), however, the other values were significantly reduced, indicating that the lordosis and pelvic tilt were less exaggerated.

The vertical line from C7 to the sacral end plate passed through the middle of the sacral end plate. There was less discrepancy in spinal alignment type between the lumbar lordosis, sacral slope values, and pelvic incidence posttreatment; thus, spinal type was classified as close to type III.

Discussion and Conclusion

This case study demonstrated the value of pelvic-spinal analysis for the measurement of sagittal alignment and for the evaluation of the effects of treatment. In this case, the treatment involved onabotulinumtoxinA injection of the hip flexor muscles along with a soft lumbar brace. Pelvic-spinal analysis showed significant improvements in sagittal alignment posttreatment.

When carrying out pelvic-spinal analysis, it is important to radiograph the whole spine and pelvis from C2 to the proximal femur using a variety of software (eg, Optispine, Keops), with the patient in standing position. Pelvic angles (pelvic incidence, pelvic tilt, and sacral slope), type of lumbar lordosis (high or low, deep or shallow), and the position of a vertical line from C7 on the sacral end plate should be measured in order to fully characterize the pelvic-spinal balance.

The authors found only 1 study in the literature that used these parameters to compare the pelvic-spinal balance of children with cerebral palsy (hemiplegic and diplegic) with healthy children.⁹ Similar to the results for L., they found that the pelvic incidence of the children with cerebral palsy was normal (46° vs 44.8° in healthy children), however, the pelvic positional parameters (pelvic tilt and sacral slope), spinal curvatures, and balance were significantly different compared to the healthy children. This suggests that the changes in spinal alignment that occur during adolescence in cerebral palsy are the result of impaired muscle tone and strength (eg, hypertonia of muscles above and below the pelvis, weakness in the posterior muscles), tendon and muscle contractures, and compensations of the skeletal system.

The parameters described in this article can be easily measured, and they characterize pelvis and spinal shape well. In a well-aligned spine and pelvis, there should be consistency between the parameters, that is, a high pelvic incidence should be associated with high sacral slope and lumbar lordosis values. If there is a mismatch between the values of pelvic incidence and sacral slope and lumbar lordosis, that is, low pelvic incidence and high sacral slope and lumbar lordosis values, this denotes a pathological alignment. This is usually caused by extrinsic factors such as spasticity or contracture or weakness of hip, pelvic, or spinal muscles that affect pelvic-spinal balance. These factors can be evaluated in the clinical assessment and appropriately treated.

Pelvic-spinal analysis is thus complementary to the clinical evaluation. It provides an objective measure of alignment. A thorough clinical evaluation is essential to determine the impairments underlying abnormal alignment. It should include spasticity, muscle strength, and active and passive range of motion. This combined approach will help to determine the role of primary, secondary, and tertiary impairments in the altered sagittal spinal alignment. Appropriate treatments can then be determined, and repeat pelvic-spinal analysis can be used to evaluate their effectiveness.

One of the goals of treatment to improve sagittal alignment is to prevent the development of pain in adulthood. Lumbar hyperextension can cause low back pain, spondylolysis, or degenerative spondylolisthesis due to stress on the posterior spinal structures. Intramuscular injections of onabotulinumtoxinA during growth may effectively restore a balanced pelvicspinal complex in ambulatory children with cerebral palsy, preventing future problems.

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Author Contributions

EC-V contributed to conception and interpretation and drafted the manuscript. J-CB contributed to analysis and interpretation and critically revised the manuscript. JD contributed to acquisition, analysis, and interpretation and critically revised the manuscript. PR contributed to interpretation and critically revised the manuscript. All authors gave final approval and agree to be accountable for all aspects of work ensuring integrity and accuracy.

Declaration of Conflicting Interests

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References

- Love SC, Novak I, Kentish M, et al; Cerebral Palsy Institute. Botulinum toxin assessment, intervention and after-care for lower limb spasticity in children with cerebral palsy: international consensus statement. *Eur J Neurol.* 2010;17(suppl 2):9-37.
- Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. *Dev Med Child Neurol.* 2013;55(10):885-910.
- Chaléat-Valayer E, Parratte B, Colin C, et al. A French observational study of botulinum toxin use in the management of children with cerebral palsy: BOTULOSCOPE. *Eur J Paediatr Neurol*. 2011;15(5):439-448.

- Molenaers G, Schorkhuber V, Fagard K, et al. Long-term use of botulinum toxin type A in children with cerebral palsy: treatment consistency. *Eur J Neurol*. 2009;13(5):421-429.
- Molenaers G, Desloovere K, Fabry G, De Cock P. The effects of quantitative gait assessment and botulinum toxin a on musculoskeletal surgery in children with cerebral palsy. *J Bone Joint Surg Am.* 2006;88(1):161-170.
- Duval-Beaupère G, Schmidt C, Cosson P. A Barycentremetric study of the sagittal shape of spine and pelvis: the conditions required for an economic standing position. *Ann Biomed Eng.* 1992;20:451-462.
- Song EW, Lenke LG, Schoenecker PL. Isolated thoracolumbar and lumbar hyperlordosis in patient with cerebral palsy. *J Spinal Disord*. 2000;13(5):455-460.
- Mac Carthy JJ, Betz RR. The relationship between tight hamstrings and lumbar hypolordosis in children with cerebral palsy. *Spine (Phila Pa 1976)*. 2000;25(2):211-213.
- Deceunink J, Bernard JC, Combey A, et al. Sagittal X rayparameters in walking or ambulating children with CP. *Ann Phys Rehabil Med.* 2013;56(2):123-133.
- Roussouly P, Gollogly S, Berthonnaud E, Dimnet J. Classification of the normal variation in the sagittal alignment of the human lumbar spine and pelvis. *Spine (Phila Pa 1976)*. 2005;30(3):346-353.
- Berthonnaud E, Dimnet J, Roussouly P, Labelle H. Analysis of the sagittal balance of the spine and pelvis using shape and orientation parameters. J Spinal Disord Tech. 2005;18(1): 40-47.
- Mac-Thiong JM, Labelle H, Berthonnaud E, Betz RR, Roussouly P. Sagittal spinopelvic balance in normal children and adolescents. *Eur Spine J.* 2007;16(2):227-234.
- Roussouly P, Gollogly S, Noseda O, Berthonnaud E, Dimnet J. The vertical projection of the sum of the ground reactive forces of a standing patient is not the same as the C7 plumb line: a radiographic study of the sagittal alignment of 153 asymptomatic volunteers. *Spine (Phila Pa 1976)*. 2006; 31(11): E320-E325.