Comprehensive assessment of therapy using the GMFM scale in a female patient with cerebral palsy treated with botulinum toxin

Ocena kompleksowego postępowania terapeutycznego za pomocą skali GMFM u pacjentki z mózgowym porażeniem dziecięcym leczonej toksyną botulinową

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Key words
spasticity, functional scale, gross motor, NDT-Bobath

Abstract
Purpose: The aim of the study was to evaluate the effects of therapy using the GMFM scale in a female patient with cerebral palsy treated with botulinum toxin.
Material and methods: The observation involved a 5-year-old girl diagnosed with spastic diplegia cerebral palsy. The study was conducted at a University Children's Hospital. The patient participated in two 3-week rehabilitation camps involving a comprehensive therapy. Each camp began two weeks after injections of botulinum toxin. The effects of the therapy were assessed using the GMFM scale, and the assessment was performed both before the treatment and after its completion.
Results: The results showed an improvement in the motor functions assessed with the GMFM scale and in the structure of the body, the body functions, and levels of activity and participation after the application of a comprehensive and systematic therapeutic intervention.
Conclusion: Regular administration of botulinum toxin alongside a comprehensive therapeutic treatment improved the patient's motor functions and enabled her to achieve new motor functions.

Słowa kluczowe
Spastyczność, skala funkcjonalna, duża motoryka, NDT-Bobath

Streszczenie
Cel: Celem pracy była ocena efektów usprawniania za pomocą skali GMFM u pacjentki z mózgowym porażeniem dziecięcym leczonej toksyną botulinową.
Materiał i metody: w obserwacji uczestniczyła 5-letnia dziewczynka, u której rozpoznano obustronne porażenie kurczowe (diplegia spastica) mózgowego porażenia dziecięcego. Badania przeprowadzono w jednym z uniwersyteckich szpitali dziecięcych. Pacjentka uczestniczyła w dwóch 3-tygodniowych turnusach rehabilitacyjnych, gdzie była objęta kompleksową terapią. Każdy turnus rozpoczynał się 2 tygodnie po podaniu toksyny botulinowej. Do oceny efektów usprawniania użyto skali GMFM, którą przeprowadzano zarówno przed terapią, jak i po jej zakończeniu.
Wyniki: Wyniki badań wykazały poprawę funkcji motorycznych ocenianych skalą GMFM, a także poprawę struktury ciała, funkcji ciała, czynności i uczestnictwa po zastosowaniu kompleksowego i systematycznego postępowania terapeutycznego.
Wnioski: Regularne podawanie toksyny botulinowej, wraz z zastosowaniem kompleksowego postępowania terapeutycznego u badanej pacjentki, wpłynęło na polepszenie i osiągnięcie nowych funkcji motorycznych.

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INTRODUCTION

Cerebral palsy (CP) is a term that commonly refers to various motor disorders, which are often accompanied by visual, hearing, speech, cognition, communication and perception impairments. Therefore, the complexity of the disorders connected with cerebral palsy requires a comprehensive therapy and a huge commitment from the patient's parents. Physiotherapy is one of the main constituents of treatments for cerebral palsy, which is why current scientific theories and technological solutions need to be followed in order to improve the patients' quality of life. Parents play a huge role in the therapy. They have to be aware themselves and to teach their child that cerebral palsy is an illness that cannot be cured entirely and requires a long-term rehabilitation. However, the comprehensive work of a therapeutic team can allow for a significant improvement in a child's quality of life.

Brain damage disorganises and delays the development of the neurological mechanisms that control posture, balance and movement. The muscles that take part in these mechanisms become inefficient, lack coordination, may have increased or decreased tension and have reduced strength. Neuromuscular motor disorders are also accompanied by musculoskeletal disorders[1]. Due to the complexity of the disorders, children with cerebral palsy reach the consecutive stages of motor development later, are less able at the consecutive stages of maturation than healthy children and have incorrect motor patterns. It should be remembered that the motor components change with a child's growth and development, and are strongly related to the child's emotional development, social adaptation, personality and intellectual capacity, as well as to the appropriate rehabilitation and the parents' care.

One of the most common challenges faced by therapists is reducing spasticity in patients with CP. Experimental studies have proven that long-term muscle tension negatively affects muscle growth, and as a consequence, it causes a reduction in the mass and length of the muscle, as well as a transformation of initially dynamic contractures into permanent, structural contractures. A local intramuscular administration of botulinum toxin type A is a modern method of reducing spasticity in patients with cerebral palsy. Administering the toxin causes the temporary chemical denervation of a skeletal muscle through blocking the release of acetylcholine, i.e. a neurotransmitter at the neuromuscular junctions. The inhibition of the neurotransmission leads to a clinical weakening and atrophy of the muscles without sensory disorders and muscle tissue necrosis[2]. This effect lasts on average from three to four months. It is believed that a local injection of the toxin can improve motor patterns or can cause new ones to appear due to the development of new branches of motor fibres within a nerve ending. The main advantage of using botulinum toxin type A is that the neurotoxic effects are reversible, and it also has a high safety profile. No serious side effects have been described so far. General weakness, fever and swelling of the injection area occur very rarely and do not last longer than two weeks[3]. It should be remembered that rehabilitation is an indispensable stage of treatment after administering the botulinum toxin. A therapist should use the period of the duration of the effect of the toxin to improve motor patterns or to teach new ones, and to monitor the effects of the treatment.

Objective ways of evaluating the effects of motor rehabilitation, which do not depend on applied therapeutic methods and the system of treatment, and which focus mainly on the patient's functionality level, have been developed and are now commonly used. One of the objective scales that allows for an assessment of the changes in the gross motor functions of a child with cerebral palsy is the Gross Motor Function Measure (GMFM) scale. This scale allows for an evaluation of the changes in functions and the determination of the level of motor development in children with cerebral palsy and Down's syndrome. The GMFM encompasses actions from activities in lying, through rolling, to walking, running and jumping.

Modern scales and classifications are designed for a detailed evaluation of the degree to which a child has mastered new skills. Using such scales shows the changes caused by therapy aimed at improving the gross motor function or independence and reducing the necessary amount of care owing to the child's increased independence.

AIM OF THE STUDY

The aim of the study was to evaluate the effects of therapy using the GMFM scale in a female patient with a cerebral palsy being treated with botulinum toxin.

MATERIALS AND METHOD

The study investigated a five-year-old girl who was diagnosed with spastic diparetic cerebral palsy with no intellectual disability. The parents granted their written consent for the family's participation in the study.

The assessment of the effects of the comprehensive rehabilitation treatment was conducted with the GMFM scale, which comprises 88 items (GMFM-88) grouped into five dimensions for evaluating a child's activities:

- Trial A, lying and rolling: evaluates 17 motor functions in a supine position and prone position, as well as evaluating the child's rolling motions;
- Trial B, sitting: evaluates 20 motor functions performed while sitting;
- Trial C, crawling and kneeling: evaluates 14 motor functions performed while on all fours;
- Trial D, walking: evaluates 13 motor functions performed while kneeling and standing;
- Trial E, walking, running and jumping: evaluates 24 functions performed while walking, ascending stairs, running and jumping.

Each motor function that a child performs is evaluated on a scale numbered from 0 to 3, where 0 denotes that a patient did not initiate a trial, 1 denotes the initiation of a trial, 2 denotes the partial completion of a trial, and 3 denotes the comple-

tion of a trial. A given trial can also be omitted; in such a case, the abbreviation 'NT' ('not tested') should be used. The child must perform each trial independently, without any help and assistance from a therapist. Once the scale has been applied, the scores are summed up in each of the five dimensions and are presented in the final result as percentage values. Furthermore, a general score is also calculated, which is a sum of the percentages from each category divided by the number of categories. In this way, a therapist can assume the activities that received lower scores or the activities that promise improvement as the aim of the therapy. The GMFM scale takes the child's achievable activities into consideration, and not the quality of their performance. It also disregards the evaluation of hand functions.

The girl was evaluated with the GMFM scale before and after the two therapy programme and again five months after the therapy's completion. In total, the patient was evaluated five times.

The study began on the day when botulinum toxin type A was administered to the patient. The dosage was 12–16 U/kg of the child's total body mass. The toxin was injected into the motor points of the ischiocrural muscles and the triceps surae muscles of both lower limbs. No undesirable effects were observed after the injection.

1st Rehabilitation camp

Two weeks after the injection of the botulinum toxin, the patient was admitted to the Rehabilitation Ward for a period of three weeks. The patient was also given short leg casts for a period of two weeks in order to increase the range of the active dorsiflexion of the ankle joints and to reduce spasticity. The patient's stay at the rehabilitation camp involved a comprehensive therapy treatment that included:

1. Motor rehabilitation according to the NDT-Bobath concept The patient's motor rehabilitation focused mainly on the activation of the trunk, the dissociation of movements and the patient's verticalisation, both in the casts and without them. Passive exercises were also used to eliminate below-knee contractures and to stretch the anatomical structures before putting on the casts and after taking the casts off.
2. Occupational therapy.
3. Classic massage.
4. Physiotherapy.

An assessment with the GMFM scale was conducted both before and after the therapy. The verticalisation of the patient at home with the use of the cast braces was also recommended.

Four months after the end of the 1st rehabilitation camp the second injection of botulinum toxin type A was administered to the patient at the motor points of the ischiocrural muscles and the triceps surae muscle of both lower limbs. The dosage was also 12–16 U/kg of the patient's total body mass. No undesirable effects were observed after the injection.

2nd Rehabilitation camp

Two weeks after the botulinum toxin was administered, the girl was admitted to the Rehabilitation Ward, again for a period of three weeks. The aim of the physiotherapy was to achieve active verticalisation and preparing the patient for independent movement in an active wheelchair. The patient was given short leg casts for two weeks in order to increase the active dorsiflexion of the ankle joints and to reduce spasticity. The course of comprehensive rehabilitation was the same as during the 1st rehabilitation camp, except for three days when the patient was not verticalised because a high muscle tension generated in the lower limbs which prevented effective verticalisation. An assessment with the GMFM scale was conducted before and after the therapy. The attending physician issued a certificate for purchasing an active wheelchair.

Five months after the end of the 2nd rehabilitation camp, the patient came to a medical appointment, during which a fifth assessment with the GMFM scale was conducted. At this time, the parents decided to end the girl's programme of comprehensive rehabilitation preceded with injections of botulinum toxin. The parents' decision was tantamount to the conclusion of the research study.

RESULTS

Figures 1 and 2 present the percentage results of the five assessments of the patient performed with the GMFM scale.

The analysis of the results, presented in a graphical form in Figure 1, shows that the motor functions evaluated with the GMFM scale in each category (A, B, C, D and E) changed as a result of the comprehensive rehabilitation. The highest percentage values, ranging between 92 and 96%, describe the motor skills that were observed in Trial A (lying and rolling) in all five assessments. The motor functions evaluated while walking, running and jumping amounted to 0% in Trial E, in all of the five conducted assessments.

For Trial A, the highest values were observed in Assessments 3 and 4, where the values equalled 96% and increased by 4% in comparison to Assessments 1 and 2. In Assessment 5, the termination of the regular administration of botulinum toxin combined with the comprehensive therapy caused a decrease in the obtained results by 4% in comparison to the results in Assessment 4.

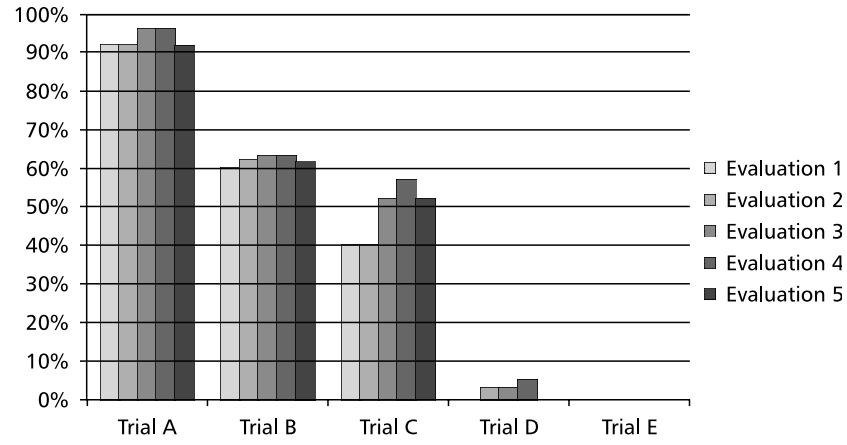
For Trial B (sitting), the highest values (63%) were also obtained in Assessments 3 and 4 which were conducted during the 2nd rehabilitation camp. These values increased by 3% in comparison with Assessment 1; and by 1% in comparison with Assessment 2. Assessment 5 showed a decrease of the values by 1% in comparison to the results obtained during Assessment 4.

Crawling and kneeling amounted to a score of 40% in Assessments 1 and 2. In Assessment 3, the study observed an increase by 12% from the values recorded in Assessments 1 and 2. The highest percentage values were obtained in Assessment 4 (57%), where the score increased by 5% in comparison to Assessment 3. In Assessment 5, the obtained results were smaller by 5% in comparison to Assessment 4.

Due to the comprehensive therapy, the percentage values in Trial D, describing motor functions performed

while kneeling and standing, increased in Assessments 2 and 3 by 3%, and in Assessment 4 by 5% in comparison to Assessment 1, where the values amounted to 0%. Assessment 5 (during the medical appointment) revealed a loss of the motor abilities acquired during the 1st and the 2nd rehabilitation camp; and the obtained values amounted to 0%.

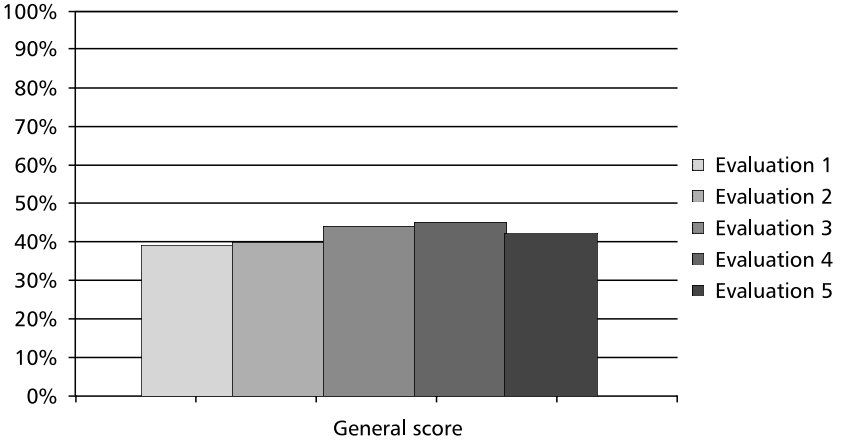
The analysis of the results, presented in a graphical form in Figure 2, shows that the sum of all motor functions evaluated with the GMFM scale changed due to the comprehensive therapy that was administered. The percentage value describing the sum of the motor skills was higher in Assessment 2 by 1% (after the 1st rehabilitation camp) in comparison to Assessment 1 conducted before the rehabilitation therapy. In Assessment 3 (before the 2nd rehabilitation camp), the percentage value describing the general score of the conducted GMFM scale was higher by 4% than in Assessment 2 (after the 1st rehabilitation camp). The highest percentage value (44%), describing the total motor skills, was observed in Assessment 4 (after the 2nd rehabilitation camp), and was higher by 1% in comparison to the value obtained in Assessment 3. Assessment 5 (during the medical appointment) showed a decrease in this value by 3% in comparison to Assessment 4. However, this decrease was not smaller than the percentage values obtained in Assessments 1 and 2.



	Evaluation 1	Evaluation 2	Evaluation 3	Evaluation 4	Evaluation 5
Trial A	92%	92%	96%	96%	92%
Trial B	60%	62%	63%	63%	62%
Trial C	40%	40%	52%	57%	52%
Trial D	0%	3%	3%	5%	0%
Trial E	0%	0%	0%	0%	0%

Figure 1

Percentage scores in each category (A, B, C, D and E) after three assessments of the patient using the GMFM scale



	Evaluation 1	Evaluation 2	Evaluation 3	Evaluation 4	Evaluation 5
General score	38%	39%	43%	44%	41%

Figure 2

General percentage results after five assessments of the patient using the GMFM scale

DISCUSSION

Cerebral palsy as a disorder may take many forms and can produce different clinical pictures, which is a result of the diversified damage that occurs in a developing brain. An important aspect in treating CP is the early diagnosis of the disorder and the beginning of long-term and systematic rehabilitation. The main cause of motor function disorders in children, which most significantly hinders their daily life and the motor rehabilitation of a patient, is spasticity. A reduction of the pathological muscle tension and introducing intensive rehabilitation can improve the quality of the functions of a given muscle and prevent contractures[2]. A modern method for reducing spasticity involves a local intramuscular injection of botulinum toxin (BTX-A).

Botulinum toxin is currently registered for the treatment of spastic pes equinus in patients with cerebral palsy in Great Britain, Europe and North

American countries, as well as Australia and New Zealand. However, it is being more and more frequently administered into muscle groups other than the ones specified in the registration materials, including muscles of the upper limbs[4].

Injecting botulinum toxin is only a pharmacological support for the rehabilitation of a patient with cerebral palsy. According to Boyd and Graham[5], using botulinum toxin type A without introducing motor rehabilitation does not cause any changes that would be noticeable six months after the injection. This is why a targeted therapy, individually adjusted for each patient, becomes a very important aspect of the treatment. Monitoring the effects of the botulinum toxin type A treatment also plays an important role. Until recently, the following measurements were used for monitoring these effects: evaluations of the range of the patient's active and passive motions; subjective measurements of the muscle tension; observational scales of gait evaluation; and three-dimensional gait analyses. Today, the results of the treatment are usually assessed functionally using gross motor function scales, such as the one applied in this study[4]. According to Molenaers et al.[6], the key factors in achieving broader functional capabilities after administering the treatment with botulinum toxin type A involve: a careful selection of the muscles intended for the injection of the medicine; using the optimal doses and injection techniques; applying an appropriate treatment before and after the injections; and administering injections with an appropriate regularity.

The duration of the botulinum toxin effect is limited. The literature most often states that its effect ends after three to four months[3]. This conclusion is consistent with the results obtained in this study, which confirm that administering botulinum toxin regularly and introducing comprehensive therapy reduced the patient's spasticity, which caused improvements in the results monitored with the functional scale. However, the evaluation of the patient after five months from the administration of the last dose of botulinum toxin showed that the spasticity had increased in comparison to the previous evaluation, which translated into worse results in the applied scale. Nevertheless, these results were not worse that the results obtained during the first and second evaluations. Depending on individual factors, the duration of the botulinum toxin effect may last for as long as six months in some patients.

In their research, Corry et al.[7] compared the effect of botulinum toxin type A injections with the effect of serial casting in the treatment of spastic pes equinus in a group of 20 children with diplegic and hemiplegic cerebral palsy. In both the group treated with the botulinum toxin and in the group treated with immobilisation, the passive dorsiflexion range of motion at the ankle joint and the contact of the foot with the ground improved after two weeks from the beginning of the therapy. However, this improvement lasted for longer in the group treated with the botulinum toxin[7]. Currently, the application of castings after the administration of botulinum toxin is considered as an important element of the therapy that can increase the patient's functional benefits. In this study, the application of short leg casts allowed for the verticalisation of the female patient and for using her arms to perform functions in a vertical position. Desloovere et al.[8] also proved that serial immobilisations after administering botulinum toxin could improve the symmetry of the pelvis. It has been suggested that this phenomenon results from the improvement of the distal stability, which then allows for a greater mobility in the proximal section[9].

Dai et al.[9] also described the administration of botulinum toxin type A alongside drugs to regulate muscle tension: Baclofen and Tizanidine. Their study encompassed 30 children with spasticity of the gastrocnemius muscle. Seventeen of the children were given Baclofen alongside an injection of the toxin; and thirteen received Tizanidine, also alongside an injection of the toxin. The effects of the treatment were evaluated with the GMFM scale and with the modified Ashworth scale. The results of the study showed a positive effect of drug combination with injection of the botulinum toxin, greater benefit has been demonstrated for the Tizanidine. No undesirable effects were noted[9].

It is believed that treatment with botulinum toxin is most beneficial for children who are able walk, have an increased muscle tension and minimal contractures, and are between two and six years of age[4]. Research conducted at the 'Górka' Hospital in Busko Zdrój encompassed 237 cases[10]. This research included patients who were injected with botulinum toxin and were between 1.5 and 44 years of age. The toxin was administered into the muscles of the lower and the upper limbs. In the study, it was observed that the effects of the botulinum toxin lasted for longer in the patients aged between three and five years than in the older patients. The aim of administering the botulinum toxin was not only to improve the patients' functions, but also to improve aspects of their aesthetics and hygiene, and to reduce the pain connected with the intensified spasticity. The observation of the effects of using botulinum toxin in the 'Górka' Hospital revealed a large degree of effectiveness in all patients who underwent the treatment[10]. Our study also obtained satisfactory treatment effects that allowed for the patient's active verticalisation, which could previously not be achieved despite the patient undergoing regular rehabilitation. The patient's verticalisation also caused an improvement in her speech and communication, as well as in the parents' levels of satisfaction.

A majority of the publications, including this paper, concern the results of the treatment in the early period after the medicine is administered. However, in a study by Maciąg-Tymecka and Sławek[2], an evaluation was conducted three years after the treatment and six months from the last injection of botulinum toxin. Their study encompassed a group of 28 children aged between two and seven years. The children were divided into Group I, which was given local injections of botulinum toxin into the triceps surae muscles and was rehabilitated systematically; and Group II,

which was not treated with botulinum toxin and was systematically rehabilitated. In Group I, after three years of the treatment, the study observed a reduction of the muscle tension in all patients, and in some it also observed an improvement of the gait pattern and gait speed. In turn, in Group II, the improvement was significantly smaller, and in the largest number of cases the study showed that movements had become restricted, deformations became permanent, and locomotor functions had deteriorated. This research did not register undesirable effects in any of the treated patients, and also observed that the patients did not develop immunity to the botulinum toxin[2].

Because almost all publications confirm the high safety record of botulinum toxin, including the present study, the toxin can be recommended for widespread use.

A good number of publications have paid particular attention to treatments with botulinum toxin type A in a population of children with cerebral palsy who are able to walk. However, the results obtained in this paper indicate satisfactory results in a patient who was unable to walk and who had a very large problem with verticalisation. More and more often, the literature is presenting observations concerning the administration of the medicine into the upper limbs. However, the evidence in favour of administering this treatment is unsatisfactory due to the limited number of controlled studies, and the lack of standardisation of the applied measurements of the treatment results[4].

The results obtained in this case study allow for the formulation of a thesis that using botulinum toxin in the female patient with cerebral palsy considerably improved her motor functions as evaluated with the GMFM scale.

CONCLUSIONS

1. The regular administration of botulinum toxin combined with the comprehensive therapy helped the child to acquire a new function, i.e. an active standing position.
2. The local administration of botulinum toxin and the introduction of motor rehabilitation improved the child's gross motor skills.
3. The termination of the regular administration of botulinum toxin and the comprehensive therapy caused a decrease and loss of certain motor functions.

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