

Review Article

Physiotherapy management of obstetric brachial plexus palsy: A systematic review of the literature

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Obstetric brachial plexus palsy (OBPP) is a pathological situation that is defined as a birth complication with the frequency of occurrence 1-3 per 1000 live births globally. The prime mechanism of injury is the complexion and/or traction of the brachial plexus and may occur in utero through the birth canal or during the process of delivery. This pathology can result in a variety of injuries with that affect the range of motion, the mobility and the use of the upper limb. Management alternatives for the treatment of OBPP consist of non-operative and operative treatment. There is a controversy regarding the selection of the appropriate rehabilitation method for the management of OBPP. The objective of the current research is the search of the literature for the current scientific evidence about the effectiveness of physiotherapy as a conservative method of treatment of infants with OBPP. According to current literature the physiotherapy methods that were used varied from passive exercise to splints and traction. There is little evidence regarding the use of physiotherapy as a primary method of conservative management of OBPP, there is need for more research to elucidate their role

Keywords: Conservative management, Obstetric brachial plexus palsy, Occupational therapy, Physiotherapy, Rehabilitation

Introduction

In a recent literature search a lack of evidence was observed concerning the effectiveness of physiotherapy management of infants that are diagnosed with Obstetric Brachial Plexus Palsy (OBPP)¹. OBPP is a complication that occurs during childbirth and the frequency of the disease in the general population is 1-3 per 1000 live births. This rate can be compared to other disorders that occur during childbirth or during the early and later stages of a child's life such as Down's Syndrome and Cerebral Palsy^{2,3}. Recovery is also reported but there is a variation to the specific rate (30-90%). The most possible explanation for the high level of the reported variation is the referral bias and the surveys that are conducted for the reports. However we should mention that nowadays it is agreed that recovery percentage is less than what was originally thought. For this reason, it is recommended that a greater number of children should be monitored and early interventions should be applied to more cases to improve the outcomes of the OBPP syndrome⁴.

Regarding the possible causes for the occurrence of OBPP there are a number of theories that assess its underlying

causes. The theory that is most accepted is that the primal cause for OBPP is the compression or a stretch injury of the whole or a part of the brachial plexus⁴. Forces that may compress or may cause stress injury are usually applied during natural birth procedure and more specifically during the fetus' passing through the birth canal. These forces could be applied laterally on the neck or as stretching forces to an isolated upper limb³. This particular mechanism results in the anterior shoulder involvement and implies a higher incidence of injuries on the left side because of the left occipital anterior being the most common presentation. These circumstances

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result in a variety of injuries such as neuropraxia and nerve root avulsion. One, several or all of the nerve roots of the brachial plexus can be affected (C5-T1 spinal location). The pathological outcomes of PBPP can be temporal dysfunction in some milder cases or functional deformity and impairment that last throughout the patient's life⁵.

The majority of the infants that are diagnosed with OBPP receive some form of treatment but there is no consensus regarding the optimal treatment method⁶. The difficulty in the selection of the most appropriate methods for OBPP treatment could be due to the observed difficulties in quantifying the recovery of the patient. There are many studies that assess the recovery of the OBPP patient and use a variety of different measures for reporting the therapy's outcome. Furthermore, there is an evident difficulty for the report of the patient's natural recovery process. There is little scientific evidence that suggests that active treatment yields better results in comparison to no treatment at all. There is, however, a general consensus that more infants diagnosed with OBPP receive some of active treatment of active treatment in the first two years of their life⁷.

The term "active treatment" that is used in a variety of cases refers to any number of interventions that can repair the brachial plexus or any form of conservative therapeutical interventions (e.g. physiotherapy). Surgical operations are usually recommended to infants that have a small percentage of improvement in the first 6 months of their life⁸. A review of the literature revealed no generally accepted criteria for application or the optimal timing for surgery⁹. This could possibly be a result of limitations in imaging techniques that currently do not accurately depict the extent of damage on the brachial plexus. Furthermore, these studies do not predict accurately the results of the expected recovery and they are unable to report small changes in the status of the injury over time¹⁰.

Objectives

The main objectives of this systematic review is to systematically search the literature and provide the current evidence for the availability and effectiveness of the physiotherapeutic interventions in infants (>2 years old) that are diagnosed with OBPP. Furthermore, the review will focus on the outcome measures of the interventions.

Methods

The studies that are included in the review are quantitative and use physiotherapy to as the primary method for conservative treatment. The studies include randomized control trials, cohort and case - cohort studies (comparative studies) and case series. The inclusion criteria further consisted of the written language (English) and the year of publication (June 2010 until July 2020). These inclusion criteria focused on discovering the most up to date relevant data on the current literature. Participants in the selected studies included infants diagnosed with OBPP

either by clinical examination or with imaging modalities and were treated by protocols that included physiotherapy in the first two years of their life. Patients that underwent primary surgery in the brachial plexus area or in the secondary deformities and patients that followed a protocol to determine the effects of a pharmacological agent (e.g. Botulinum toxin) were excluded.

The primary outcome measure of the selected research papers was the patient's recovery. There are a variety of measures that estimate the patient's recovery process such as strength, range of motion of the affected limb and the overall sensation of the area^{7,10}. There are a number of researches that use specific instruments to measure the patient's recovery (inclinometer) but there was no general consensus for the use of certain instrument.

Prior to the database search we conducted a small scale search in the same databases to ensure that there is no similar review published before or currently being reviewed. Our research found no prior systematical review of the literature.

The research strategy was systematical, and it can be reproduced. Overall, 5 databases were searched to identify a number of studies that fit to our criteria of selection. The databases included MEDLINE, CINAHL, Science Direct, Physiotherapy Evidence Database (PEDro) and Evidence Based Medicine Reviews. Due to the similarity of the indexing terms in the databases a common search algorithm was developed for all databases. The search strategy was designed according to the methodology described by Brettle et al.¹⁴.

The search algorithm that was used included: "brachial plexus neuropathies" OR "birth injuries" OR "exercise movement techniques" OR "physiotherapy" OR "rehabilitation" OR "treatment outcome".

Methodological Quality of the Studies

The studies that were obtained from the initial searching algorithm were assessed independently to assure that the inclusion criteria are met and more specifically to assess the type of study, the patients that participated in the research and the outcome measures.

Each of the studies that met the inclusion criteria was critically appraised for its methodological quality using the Critical Review Form for Qualitative studies as was proposed by Law et al.²⁵ (Appendix I). This particular qualitative appraisal tool is design to examine the internal and external validity of the study and its outcome measures correspondingly. To provide an overall quality overview of each study the Critical Review Form tool provides a score system with two options, 0 and 1 where 0 does not fulfill the specified criterion and 1 completely fulfills the specified criterion. There are a number of 16 questions and an overall score is provided to indicate the study's quality with 16 being the greatest quality score.

In the discussion section, a description was included of the

selected studies, regarding the method of diagnosis of the OBPP and the patients that were included, the intervention of the studies and control groups and the outcome measures (type used and time). Finally, an effort was made to determine the clinical homogeneity of the studies by examining the included participants, the interventions and the outcome measures. In cases of heterogeneity there were appropriate clarifications in the text.

Results

The search strategy provided a sum of 411 initial studies. The next step was to remove any duplicate studies but also to exclude the studies that did not meet the inclusion criteria. This process yielded a total of 24 studies that were reviewed in their full text version. After the review, 16 of the 24 articles were discarded because they did not meet the inclusion criteria and the systematic review was conducted by the analysis of 8 studies²¹⁻²⁹. The first article, written by Socolonsky et al., 2016. Two principle treatment procedures have been utilized: essential medical procedure, comprising in investigating and recreating the influenced segments of the brachial plexus inside the initial not many months of the patient's life, what's more, auxiliary systems that incorporate ligament or muscle transfers, osteotomies, and other muscular strategies. Auxiliary systems should be possible as the lone careful treatment of OBPP or after essential medical procedure, to limit any residual deficiencies. Neurolysis, nerve transfer or reconstruction with crafts are the main surgical procedures that can take place in the treatment of OBPP. Significant distributed proof accumulated over many years of careful experience favours essential nerve surgery as the underlying helpful advance in patients who don't recuperate unexpectedly, trailed by optional medical procedures for additional utilitarian improvement. As depicted in this survey, the aftereffects of such treatment can significantly improve work in influenced appendages. For best outcomes, multi-disciplinary groups should treat these patients.

In the article written by Marcus & Clark, selection criteria are considered helpful. Determining the seriousness of a solitary harmed fringe nerve at the hour of introduction is adequately troublesome; the test is far more noteworthy when the variable injury includes an unpredictably designed what's more, not entirely comprehended gathering of nerves. The board of obstetrical sores of the brachial plexus in the infant is rich with disagreeing conclusions. Microsurgical treatment of obstetrical brachial plexus injuries is a moderately youthful field of careful skill. Specialists occupied with the early exertion to improve useful result for newborn children with this conceivably deep rooted hindrance immediately perceived the need to set up suitable careful rules. The assurance of careful signs proceeds to be the most significant, and generally troublesome, thought for those really focusing on patients and families confronted with the physical and passionate effect of this condition.

In the study by Jeevannavar et al., 2020, 28 children were included in a research. The study showed that boys are more likely to suffer by BPPP. The study also showed that the vast majority of the children that suffer from the disease, are delivered with natural delivery and that 40% of them are delivered with an assistant instrument, i.e. a vacuum. In the study of DeFrancesco et al., 2017, the researchers analysed the database of children with BPPP, from 1997-2017. Over a 16-year time span, the frequency of BPBP fell drastically, resembled by a huge expansion in the pace of cesarean conveyance. Foundational changes in obstetric practice may have added to these patterns. As the greater part of BPBP cases have no recognisable danger factor, imminent examination of set up danger elements and portrayal of new infection determinants are expected to all the more dependably recognise babies at most serious danger. Racial and geographic disparities in sickness weight ought to be explored to distinguish interventional targets.

Lastly, in the study of O'Berry et al., 2017, brachial plexus wounds in the infant are moderately normal and in all probability, pediatricians will experience influenced babies and youngsters in their practices. The board of children with brachial plexus wounds will fluctuate as per the level of nerve injury and furthermore, inclusion. Rehabilitation techniques that are used in this study are kinesiotaping and bracing the surgical wound. A multidisciplinary approach that fuses restoration methodologies and careful intercessions can assist with enhancing recuperation and forestall long haul hindrance.

Critical evaluation

Regarding the type of study in the included papers, 3 of the studies were comparative and more specifically they compared the results of conservative treatment versus surgical treatment options^{23,27,28}. The surgery procedure consisted involved internal neurolysis and neuroma excision and nerve grafting or nerve transfer. The aforementioned studies did not include any randomisation process in the infants that consisted the treatment groups and therefore they are characterised as III-2 by the NHMCR Hierarchy of Evidence. The 5 remaining studies^{21,22,24-26} were case series and are classified as Level IV. In these five studies the overall functionality of the affected limbs were examined before and after the conservative management interventions but we must underline that there was no comparison between conservative interventions and any other form of therapy. Both comparative and case series studies have bias due to their design and it must be taken into account before the interpretation of any results.

Quality of the methodology in the selected studies

The average score of the methodological quality of the studies was 9.2, with the highest score being

11 in the research by Strombeck et al., and the lowest score 7 in the study by Leblecioglu et al. All studies described the participants sample in detail, especially regarding the demographic characteristics of the infants that were involved in the research. However, there was not a thorough description of the diagnostic criteria for the OBPP. The authors of all the studies discussed the results according to their clinical relevance and their implementation in clinical practice.

Most of the authors²¹⁻²⁵ justified their selection of the sample size, however this justification was based on circumstantial terms (number of patients in a certain time period) and not on any statistical methodology.

All the researches included biases and more specifically these biases were selection bias (infants undergoing surgical treatment were more severe cases of OBPP compared to infants that underwent conservative treatment), interventions (some of the infants that underwent surgery also received physiotherapy) and scientist bias (scientists that participated in the investigation had prior knowledge of the infants included in the study groups). It is also of great importance to mention that none of the reported results were accurately described in a methodology that allow for accurate replication. Furthermore, the outcome measures that were used by the authors to evaluate the outcomes of the interventions were not used to assess the change of the patient's status over time. Less than 50% of the selected studies analyzed their data statistically with the goal of finding any statistical significance and the same percentage analyzed in detail the limitation of their study. For the above reasons it is clear that the interpretation of the results should be done with appropriate caution.

All participants received some form of physiotherapy treatment in the first 2 years after their diagnosis of OBPP. The conservative therapeutic intervention included home exercise programs^{24,27}, mobilization programs (active and passive movement)¹⁸, mild regular exercise²³, exercises that focus on strengthening and development of the affected muscle groups²², splints²² and dynamic traction²⁵. Furthermore, 4 researches did not describe in detail the therapeutic interventions used but mentioned them as physiotherapy^{23,26}, conservative treatment^{27,28} and occupational therapy²⁶. All of these researches did not use any information about the specific exercises that were used in the protocol (duration of the exercises, frequency, and number of repetitions) but all the interventions were of physiotherapeutical nature.

Intervention outcomes

The reported recovery rates for the conservative treatment approaches ranged from 17%²³ to 100%²³⁻²⁴ and the majority of the studies mentioned a full or nearly full recovery of motion in approximately 80% of the sample^{21,23-26,28}. The evidence about the comparison between physiotherapy and surgical treatment were conflicting. For

instance in the study by Laurent et al., there was reported a 100% of recovery in children that received surgical treatment in comparison to 90% of recovery in children that received some form of physiotherapeutic intervention. In this particular research the outcome measures were the function of the affected muscle groups (deltoid, biceps and triceps) with the British Medical Research Council Muscle Movement Scale²⁹. On the other hand, in the research by Strombeck et al.²⁷, the infants that received surgical treatment had 30% more active shoulder movement in compared to infants that received physiotherapy. The range of motion was categorised with a scale 0-3 with 0 reflecting no movement and 3 reflecting normal movement. The score for the range of motion was for five directions of movement (abduction, flexion, extension, internal and external rotation). Similar findings were reported by Xu et al.²⁸ that reported a 70% of recovery in infants treated surgically (nerve transfer or nerve grafting) in comparison to the group of infants that underwent physiotherapy treatment. The results were quantified using the Mallet Scale (five point scale) that measures the infant's ability to perform a number of upper limb movements (shoulder abduction, hand to mouth, external rotation, hand to nape of the neck and hand behind back)³⁰.

Seven of the eight researches that were included in this review used outcome measure tools that were specifically designed for the infants with OBPP or with older patients with pathologies of the peripheral nervous system²¹⁻²⁶. The tools that were used to assess the outcomes of the interventions were dependent on the variables that were recorded (muscle strength, active movement) but there were cases that reported a total score as a combination of the above variables). The area of assessment was also of great significance for the selection of the appropriate measurement tool (upper limb muscle groups or whole upper limb).

The majority of the studies used either the British Medical Research Council's Muscle Movement Scale^{23,25} or the Mallet Scale^{21,26,28}. Nonetheless, there are a number of shortcomings in the use of these scales the most prominent of which is that the Mallet scale is usually employed for children that are approximately 4 years old or older (they can follow a set of instructions). The researches by Bisinella et al., and Leblecioglu et al., used a sample that was of lower age (infants) and there was not a precise description about any scaling that was followed^{21,26}. The British Medical Research Council Muscle Movement Scale was used in the studies by Lauren et al., and Linden et al., but they were used to measure the functionality of different muscle groups. For this reason it is difficult to use the results of this particular scale as combined numbers in a meta - analysis.

In the research by Stromberck et al., a special mentioned was made about the impairment that usually observed in clinical practice in children with OBPP. These impairments encompassed grip strength, manual activities in both upper limbs, joint movement and sensibility during palpation²⁷.

A lack of accuracy was also observed in the time when the

outcomes were measured. The time of assessment ranged from 12 months²³ to 5 years²⁷ depending on the research protocol. The exact time of the assessment was not reported in detail^{21,24,26,27} or was neglected in the final report in the publication. The most detailed description was reported in the research by Lauren et al., that timed the outcome measures in 2, 4 9 and 12 months of the infant's age²³. Time plays an important factor in the progress of the disease and infants are reported to clinically improve over time although the process can be long and slow³¹.

Discussion

This review aims at analyzing the effects of physiotherapy treatment (a conservative method) for the management of the OBPP. The strategy plan that was followed during the search in the literature databases was systematic and secondary searches were also made to discover possible unpublished evidence^{32,33}. The research strategy included researches that were published in the English language and date restrictions were set because recent studies about systematic reviews suggest that the methodology of studies increase over time, especially since 2000³⁴. We must also mention that due to the heterogeneity of the outcome measures the results of the studies cannot be combined in a meta - analysis.

The main goals of a physiotherapy program is to prevent the shrinkage of the antagonist muscles and the maintenance of the flexibility in the soft tissues that surround the affected muscles. In a case of nerve paralysis the affected muscles atrophy and lax and finally overextend. At the same time the unaffected muscles, with antagonize the movements, contract and shrink. This results in the loss of flexibility on the joints and at length causes malformations in the affected joints. A well designed physiotherapy program can also treat vascular disorders (oedema, hematoma) but can also train the patient in motion control by making use of the movement patterns that are already in existence. This can lead to the prevention of pathological standard adoption. On the other hand, physiotherapy can also contribute in the parent's training and help them understand the child's situation so that they will shape the environment appropriately¹¹⁻¹³.

In order to create the optimal program the physiotherapist need an initial evaluation of the affected infant's condition. The goal of the initial evaluation is to gather information about for the design of the optimal treatment approach for each case. However, the evaluation does not stop at this point but continues and physiotherapists re-evaluate the infant's condition and examine the progress that has been made. The goals of physiotherapeutic evaluation in the case of an infant with OBPP are:

- diagnosis of the pathological condition,
- the detailed analysis of the function as an important phase of the patient's rehabilitation,
- the evaluation and the accounting of the progress¹⁴.

At the same time the evaluation assess the motor function and tries to obtain a comprehensive picture of the patient's condition, determine the cause of the dysfunctions and try to predict a number of problems that may appear in the future. The physiotherapist must have knowledge about the muscle synergies that affect everyday activities in order to create a profile of the muscle activity by observing muscle contraction and muscle function, by comparing the observation to healthy individuals at the same age and by evaluating passive movement to measure muscle length and flexibility¹⁵.

When the physiotherapist makes a final decision on the appropriate program for a patient he must take into consideration a number of parameters. More specifically age is a very important factor for the selection of the appropriate exercises. Moreover, the patient's condition is contributes to the choice of the appropriate program. When the overall patient's condition improves, the rehabilitation program becomes more complex while stagnation in progress redefines the rehabilitation's needs¹⁵.

A number of physiotherapeutic techniques are used for the rehabilitation of infants with OBPP. Splints can be applied since the first day of the diagnosis of OBPP. The appropriate application of the splint contributes in the safe immobilisation of the affected member. This immobilisation can lead to the faster oedema absorption and the creation of the appropriate conditions to regenerate the affected nerve. However, the use of a splint is not recommended in some cases¹⁶. For instance, in the research by Schenk et al., it was reported that splints were the cause for a delay in the participant's rehabilitation¹⁷.

Electrotherapy is considered one of the most important physiotherapeutic techniques and is used in the treatment of OBPP. More specifically, it is the selected rehabilitation method in cases where no passive movement is detected. The selection of electrotherapy helps in the maintenance of the best possible condition in muscles with electrical stimulation by providing the minimum stimulation that is absent in OBPP. The main goals of the electrotherapy technique is to maintain the contracting muscle elements, to delay muscle atrophy, to help in the continuous blood supply of the muscle and to constantly stimulate the muscle so to avoid the complete degeneration. The process of electrical stimulation includes 15-20 contractions for each muscle group for 2-3 repetitions in each session of physiotherapy¹².

In kinesiotherapy a number of passive movements is applied to maintain the affected muscle's elasticity, prevent the adhesion and strengthen the circulation. The frequency of the passive movements is 2-3 times every day for 10 minutes. During the application of this method, the splint is removed, however to avoid the false mobility in the affected member the central part of the joint is immobilised. Regarding the degree of difficulty of passive movements, it is gradually increased to avoid creating a feeling of discomfort in the patient. Passive movement rehabilitation programs are

applied several times per day and this creates the necessity for the parents to become familiar with the technique¹⁷.

The most important cause for the development of malformations is the disturbance of the muscle equilibrium. Stretching is proven to aid the muscles in maintaining their elasticity and prevent the malformation of the affected limbs. By executing a protocol of stretching the patient elongate the muscles beyond their length in a relaxed state. The movement's rate is slow to avoid the activation of the myotatic reflection and the contraction of the muscle. Furthermore, in the case of fast movement, collagen between tissues provides passive resistance¹⁸.

When the physiotherapist has knowledge about the normal development of the infant's motor behaviour they have the ability to modify the rehabilitation program accordingly. Active movements are included in the therapeutic program when the infant is at least 5-6 months of age when the physiotherapist can take advantage of the extensive muscle activity. The success of the active movement's implementation is based on the right choice of movements according to the child's limitations and interests. When the movements patterns resemble a game physiotherapists can pick the infant's interest. The initial movement application is performed with the physiotherapist's help but as the infant develops the movements adjust to the progress¹⁹.

In the case of hydrotherapy mild massages is applied with the help of water and at the same time the water's temperature and buoyancy help in the recovery process. This physiotherapy method is very effective, especially in the early stages of rehabilitation because a substantial amount of movements are performed in the water. In more advanced stages of rehabilitation, hydrotherapy can help in muscle strengthening²⁰.

The majority of the infants affected with OBPP receive mainly conservative management as the primary forms of treatment, with physiotherapy and occupational therapy the main conservative interventions that are used. The aforementioned therapies focus on the early mobilisation and facilitation of the limb mainly affected by the pathology of OBPP and aim at preventing complications due to immobility (contraction of soft tissues, deformities of the joints). A number of examples of primary methods for conservative management are electrotherapy, functional exercise and splinting^{21,22}. However, there is no consensus regarding the efficiency of these interventions. In the following paragraphs we try to provide the latest evidence regarding the use of physiotherapeutic interventions in OBPP to determine whether these methods are efficient according to the reported evidence¹³.

This review showed that all forms of physiotherapy that were used in this review were characterised as primary conservative management in infants diagnosed with OBPP. The main physiotherapeutic assessments that were used are active range of movement correction²⁵, developmental exercises²⁴, strengthening exercises²⁴, gentle exercise in a

regular basis to prevent rigid abnormalities²⁰. However, all of the studies that we included in this review were unclear about the exact description of the protocol and for this reason the exact replication of the intervention is difficult. Physiotherapy treatments combined a number of exercises in a single session^{35,36}.

Conclusions

Physiotherapy is well established as a primary conservative method in the management of OBPP. This rehabilitation practice has the main goal to facilitate active movement patterns and strengthen the affected upper limb. At the same time patients that are subjected to physiotherapy tend to have less joint contractures and deformities in the secondary joints. However, we must underline that even though this practice is being used in the typical conservative management of an OBPP patient in many rehabilitation centers there is little evidence in research that supports this fact. In the literature review that was conducted a small number of studies were found in the initial criteria in contrast to the research conducted in other rehabilitation practices for OBPP (surgical interventions etc). This is the main reason that prevents us from drawing conclusions regarding the effectiveness of physiotherapy regarding the measured outcomes in infant rehabilitation. Furthermore, the papers that were included in the research had low methodological quality and lack in psychometric properties, variable assessment times and lack of comparison between groups of participants. Thus, clinicians cannot extract guidance from evidence regarding the results of physiotherapy intervention.

The above considerations further underline the need for research to determine the results of conservative management in infants with OBPP. To achieve scientific justification outcome measures with scientific credibility should be used to accurately monitor the patient's progress over time. The outcome measures should also monitor variables of interest or importance to clinicians but also to the families with children diagnosed with OBPP. It is clear that future research should focus on the development of new and accurate outcome instruments with accurate scientific properties that focus on the population.

The review of the included studies also led us to the conclusion that most researches were conducted by surgeons or physicians with small participation of physiotherapists. This is a major drawback in the field of the research because OBPP is a disease that should be approached multidisciplinary with the participation of physiotherapist or occupational therapies in the overall management of the patient's health.

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