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REVIEW

The effectiveness of allied health care in patients with ataxia: a systematic review

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Abstract Many patients with cerebellar ataxia have serious disabilities in daily life, while pharmacological treatment options are absent. Therefore, allied health care is considered to be important in the management of these patients. The goal of this review is to evaluate scientific evidence for allied health care in cerebellar ataxia, to identify effective treatment strategies, and to give recommendations for clinical practice and further research. A systematic search for clinical trials concerning allied health care in cerebellar ataxias was conducted using the electronic databases of PubMed, Medline, Embase, Cinahl and Pedro, and references lists of articles, in the time period from 1980 up to and including December 2011 in English and Dutch. We identified 14 trials, of which the four best studies were formally of moderate methodological quality. There was a wide variation in disease entities and interventions. The combined data indicate that physical therapy may lead to an improvement of ataxia symptoms and daily life functions in patients with degenerative cerebellar ataxia (level 2), and in other diseases causing cerebellar ataxia (level 3). When added to physical therapy, occupational therapy might improve global functional status, and occupational therapy alone may diminish symptoms of depression (level 3). There are insufficient data for speech and language therapy. Despite the widespread use of allied health care interventions in cerebellar ataxia, there is a lack of good quality studies that have evaluated such interventions. We found some support for the implementation of physical therapy and occupational therapy, but more research is needed to develop recommendations for clinical practice.

Keywords Ataxia · Cerebellar ataxia · Allied health care · Physical therapy · Systematic review

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Introduction

Ataxia is a neurological symptom that is characterized by loss of coordination of movements. The term cerebellar ataxia is used to indicate ataxia that is due to dysfunction of the cerebellum, which leads to disturbances in gait, balance and coordination, dexterity, eye movements, and speech. Ataxic gait is associated with falls and limitations in daily life [12, 16, 31, 40]. Despite the known impairments in practice-dependent and adaption motor learning [12, 35, 36, 39, 45, 51, 55], patients with damage of the cerebellum are capable of learning new strategies and to compensate for the deficits [31, 35, 37].

There are many causes for cerebellar ataxia, but a challenging group are the degenerative cerebellar ataxias. These are progressive, mostly untreatable disorders and lead to serious deterioration of mobility, independence, and quality of life [7, 48]. For many patients with ataxia, regardless of the cause, there are no pharmacological treatment options. Patients are often referred for allied health care interventions, which are considered the cornerstone in further management. Physical therapy, speech therapy, and occupational therapy are expected to prevent secondary complications and minimize dependency in daily life in patients with ataxia. However, despite the common use of allied health care, there is an unclear scientific status with regard to the effectiveness of these interventions and a lack of evidence-based treatment guidelines. This obviously results in a marked heterogeneity of treatments that patients with this movement disorder receive.

As a first step towards a more evidence-based practice, we systematically evaluated the evidence for allied health care interventions in cerebellar ataxia.

Methods

Search strategy

A systematic search was performed in the electronic databases of PubMed, Medline, Embase, Cinahl and Pedro. Clinical trials were identified using a combination of the following terms and MeSH terms: cerebellar ataxia, ataxia, physiotherapy, physical therapy, training, exercise, rehabilitation, allied health care, speech therapy, language therapy, voice therapy, and occupational therapy. The selected time period was January 1, 1980 to December 31, 2011, and the articles had to be published in English or Dutch. The retrieved articles were examined for useful references.

Selection

Articles were included if they were prospective clinical trials evaluating the effectiveness of an allied health care intervention (i.e. physical therapy, speech and language therapy, or occupational therapy) in patients with cerebellar ataxia, without interfering co-morbidity. The interventions and outcome measures had to be clearly defined in the article. Studies assessing patients both with and without cerebellar ataxia were only included if individual data for the patients with cerebellar ataxia could be extracted. Case reports or series were considered if at least two different studies described the same treatment methods.

Evidence grading

The selected articles were assessed by two of the authors (EF, SK) and the findings of the studies were extracted and summarized. Both the quality of the study design and the conclusions were appraised according to the classification of the levels of evidence using the EBRO classification (Table 1) of the Dutch Cochrane Centre and the Dutch Institute for Healthcare Improvement (CBO), member of the Guidelines International Network (GIN). [8] In case of disagreement, a third author (BvdW) was consulted to reach consensus.

Because of a substantial heterogeneity between the included studies concerning disease entities, treatment goals, interventions, follow-up period, and outcome measures, a meta-analysis could not be performed.

Results

An overview of the literature search is shown in Fig. 1. Up to 409 references per database search were found, of which 33 articles were selected. In addition, five articles were identified through reference lists. Two articles described the same study, and were considered as one publication [26, 27]. Of these selected 37 publications, 11 were excluded because of the pre-defined selection criteria. Four studies were excluded because of a retrospective study design [6, 19, 20, 30], three due to possible interfering co-morbidity [5, 46, 52], and one for not describing which interventions were used [50]. One study was excluded since the effect of the intervention was only observed at baseline; the follow up period was evaluated by telephone, with no monitoring of the activities of the participants [15]. We were not able to obtain the full text of two articles [29, 41]. Of the remaining 26 studies, three did not involve training, but focused solely on a purely measurable

Table 1 The EBRO classification

Classification of methodological quality of individual studies	
A1	Systematic review of at least two independently conducted clinical studies at quality level A2
A2	Randomized double-blind controlled studies of good methodological quality, sufficient power and consistency
B	Comparative studies, not reaching the criteria for A2, including studies with retrospective or patient-control group study design
C	Non-comparative study design/patient series
D	Expert opinion
Classification of the level of scientific evidence	
1	Conclusion based on a systematic review at quality level A1 or at least two independently conducted clinical studies at quality level A2
2	Conclusion based on a quality level A2 study or at least two independently conducted clinical studies at quality level B
3	Conclusion based on a study at quality level B or C
4	Conclusion based on expert opinion

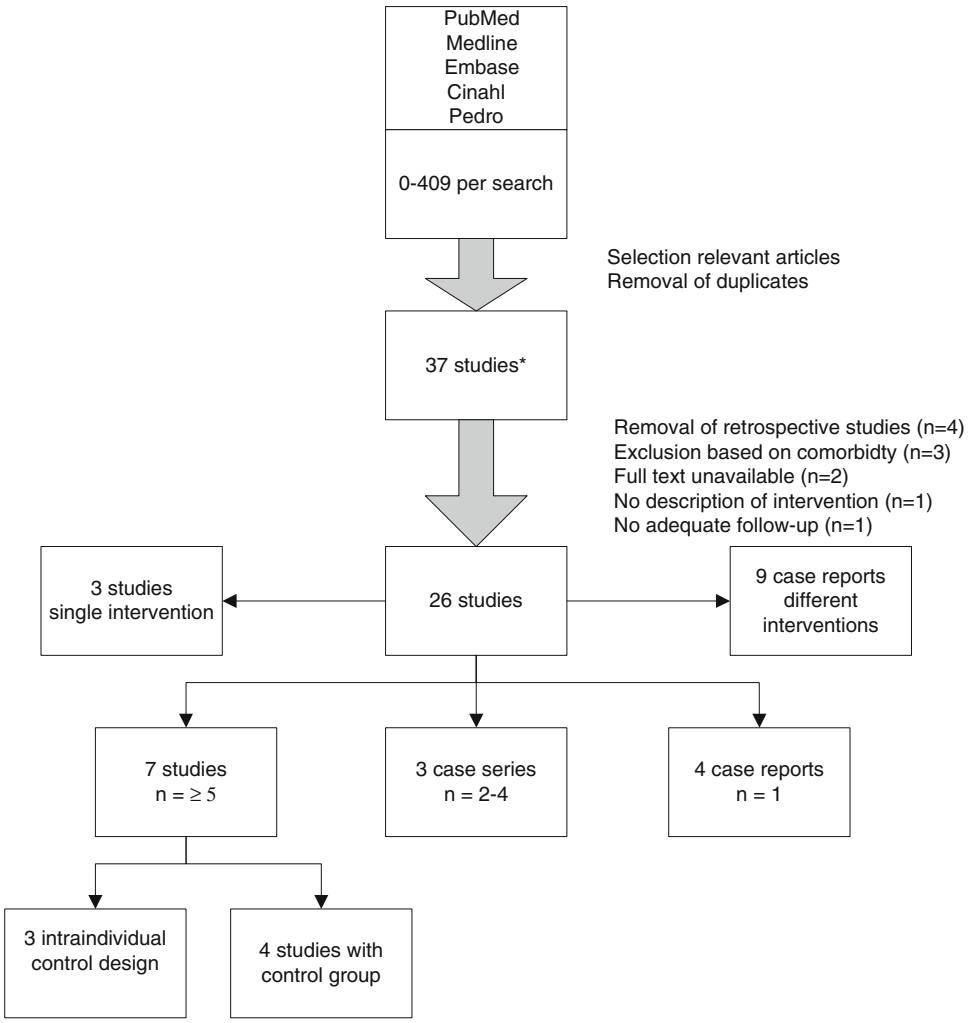

Fig. 1 Flow diagram of literature search. *38 studies, two considered as one, because they were of the same study project

kinematic effect of single interventions, and were excluded for that reason [4, 10, 43]. Of the 13 selected case reports [1, 9, 13, 14, 18, 21–25, 32, 44, 47], only four could be paired because of similar treatment methods [9, 18, 23, 24].

Finally, 14 studies remained, of which seven studies had five or more subjects with cerebellar ataxia [2, 3, 26–28, 38, 49, 54]. Only four studies could be classified as level B methodological quality, i.e. they included a control group

instead of only using within-patient comparisons [2, 28, 38, 54]. Of these four studies, three used randomization either for the whole or for a part of the intervention [2, 38, 54] and three involved a blinded assessor [2, 28, 38]. A summary of the selected articles with at least five subjects is given in a supplementary Table, Online Resource 1. The included studies were heterogeneous with respect to diagnosis, interventions and outcomes. The interventions focused on several cerebellar impairments and limitations in daily life, and varied in frequency, duration and types of exercises.

Physical therapy

Thirteen studies evaluated the effects of physical therapy [2, 3, 9, 11, 15, 18, 23, 24, 26–28, 34, 38, 53, 54], of which two were in combination with occupational therapy [28, 38]. Of these, six included at least five subjects [2, 3, 15, 26–28, 38, 54]. Four of these studies could be classified as methodological quality level B (Online Resource 1) [2, 28, 38, 54]. Physical therapy often focused on more than one domain, e.g. gait, balance, coordination, posture and muscle strengthening. Interventions applied consisted of conventional physical therapy exercises, computer assisted training, treadmill training, and biofeedback therapy.

Conventional physical therapy exercises

Conventional physical therapy targets at least one of the following domains: balance, gait, coordination, strength, endurance, and posture. In the majority of physical therapy interventions, aids (e.g. a cane or a walking frame) and physical therapy equipment (e.g. a balance ball, weights or a treadmill) were used.

In a study combining physical therapy with occupational therapy in 42 patients with degenerative cerebellar ataxia, physical therapy, with a focus on balance, gait, general condition, muscle strength and range of motion, led to a reduction of ataxia severity and fall frequency, and an improvement of gait speed and activities of daily living (ADL), compared to half of the patients ($n = 21$) who received the same treatment 4 weeks later. The improvement was more prominent in trunk ataxia than in limb ataxia. Within-patient comparisons were used to analyse long-term effects, and showed that patients with mild ataxia severity experienced a more sustained improvement. In more than half of the participants, improvement of at least one item was maintained after half a year [38]. Another study in patients with degenerative cerebellar ataxia, including ten patients with predominant effects on the cerebellum and six patients with predominant afferent ataxia, used coordinative training with the main goal of being able to activate and engage control mechanisms for balance control and multi-joint coordination. Significant improvements in ADL and in gait and balance parameters, as well as a reduction of ataxia severity were seen after 4 weeks of training, compared to baseline. Training effects were more distinct for patients with intact afferent pathways. Improvements in motor and ADL performance persisted after 1 year and seemed to be influenced by training intensity at home [26, 27]. Training balance, gait, and muscle strength in five patients with chronic ataxia due to traumatic brain injury, led to an improvement in functional independence for gait [3].

A study was conducted in 26 patients with ataxia due to multiple sclerosis (MS), combined coordination exercises, gait training, balance training, and vestibular exercises. Significant differences were found in sensory test scores, anterior balance, gait parameters, diadochokinesis, equilibrium coordination tests, and Expanded Disability Status Scale (EDSS), compared to baseline. Limb ataxia was more resistant to physical therapy than trunk ataxia. Use of Johnstone Pressure Splints in addition to exercise was of no further benefit [2].

One study in 37 MS patients with ataxia found that physical therapy with weighting, together with occupational therapy, resulted in improvement of functional ability concerning ADL tasks, fatigue, and physical functioning, compared to patients ($n = 9$) who did not receive any intervention. The physical therapy was focused on promoting normal posture and movement using weight bearing, damping and weighting, joint approximation and compression, and stimulating automatic equilibrium reactions using a gymnastic ball [28].

Treadmill training

Treadmill training in two patients with ataxia due to traumatic brain injury improved gait and balance parameters [53]. Locomotor training using bodyweight support on a treadmill and over-ground walking in a patient with traumatic brain injury lead to improvement of balance, gait parameters, motor tasks and isometric trunk endurance tests. [18] In a child with severe ataxia after a cerebellar infarct, bodyweight support on a treadmill and during over-ground walking improved her walking and transfer abilities. [9].

Relaxation and biofeedback therapy

Three articles suggested a positive effect of biofeedback therapy in patients with ataxia. [11, 23, 24] In case series using electromyogram (EMG) biofeedback in three patients with ataxia due to multiple sclerosis and degenerative cerebellar ataxia, subjects were able to reduce the

amount of inappropriate coactivation of muscles groups [11]. Relaxation and EMG biofeedback decreased severity of ataxic tremor in two case reports with patients with traumatic brain injury [23, 24].

Computer-assisted training

Improved coordination of the upper limbs after training with adaptive robot therapy was suggested in a study of eight MS patients with ataxia of the upper limbs. The training consisted of performing planar reaching movements while grasping a handle of a robotic manipulandum, which generated forces that either reduced or enhanced the curvature of the movements [54].

Supervised sports

One study noticed that patients showed improvement of velocity and speed symmetry in pointing movements of limbs, balance and manual dexterity after climbing training. The four participants had upper and lower limb ataxia of different acquired causes [34].

Occupational therapy

One study assessed the use of occupational therapy for cerebellar ataxia [49]. Another two evaluated its use combined with physical therapy [28, 38]. In one study, 26 patients with SCA3 were treated with occupational therapy alone, using the rehabilitative compensatory model [17]. Hamilton scores for depression improved, but disability scores and quality of life scores remained stable. Therapy was adjusted to the needs of the patients [49].

Occupational therapy, combined with physical therapy, including the provision of equipment and advice on ADL tasks, led to a significant improvement in speed and ability of completing ADL tasks in a study with 37 patients with ataxia due to multiple sclerosis [28].

Intensive rehabilitation with occupational therapy combined with physical therapy was shown to be of benefit in a study of 42 patients with degenerative cerebellar ataxia. Occupational therapy focused on ADL tasks, relaxation, personal hygiene, but also on balance exercises, coordinative tasks of the upper limbs and trunk, and dual motor tasks. Positive effects on functional independence, gait, ataxia severity and falls were found, and maintained in more than half of the participants after half a year [38].

Speech and language therapy

Only two case reports described the use of speech and language therapy, but were not included [44, 47].

Conclusions

Table 2 contains the level of scientific evidence for the various interventions and shows the etiological category for which these interventions are applicable. We were not able to make more than two conclusions at level 2 of the EBRO classification, meaning they were based on at least two independently conducted comparative studies, but not randomized, not double-blind controlled, or with insufficient power and consistency (Table 1).

Discussion

In a Spanish study of degenerative cerebellar ataxias, the costs for health care and society were estimated to be around 24,500 USD per patient per year. The study showed that a substantial part of the direct healthcare costs was spent on allied health care [33]. Apparently, patients with functional impairments due to ataxia are often referred to allied health care workers and these referrals have become a sort of routine in the management of these patients. While an effect of such interventions is anticipated, and already experienced as such by healthcare workers and patients alike in daily practice, there is a lack of good quality clinical trials of the effects of allied health care in patients with cerebellar ataxia.

Of all allied health disciplines, the efficacy of physical therapy has been evaluated most. This is in line with other neurologic conditions, such as stroke and Parkinson's disease. Given that balance problems are a hallmark for ataxia, it was expected that for these disciplines the most evidence would be found. Most of the studies identified and graded were case studies, with a moderate to poor quality of methodology. Moreover, there was a wide variation in disease entities, sample sizes, types of intervention, and outcome measures. The duration of the follow-up was usually short and variable. Based on the relatively poor quality of the studies that passed our inclusion criteria and that we have thus reviewed here, we re-examined the retrospective studies we had excluded. However, this provided no further insight, since they dealt with immediate stroke recovery ($n = 2$), involved a non-specified intervention ($n = 1$), or included an insufficiently characterized patient group ($n = 1$) [6, 19, 20, 30].

In general, therapy was tailored to the individual, which is in agreement with therapeutic principles, but makes it difficult to generalize these treatment protocols. We have here lumped the various aetiologies of cerebellar ataxias together, but we appreciate that aspects such as comorbid feature, or the intrinsic capacity to improve will differ per disease. In most trials no control intervention was used, which complicates the interpretation of whether the

Table 2 Summary of the level of scientific evidence for the various interventions

Conclusions	Diagnosis	Level of scientific evidence
Physical therapy		
Conventional physical therapy		
It is plausible that physical therapy improves ataxia severity, balance, gait, fall frequency and ADL functioning [26, 27, 38]	Degenerative cerebellar ataxia	2
Limb ataxia seems to be more resistant to physical therapy compared to trunk ataxia [2, 38]	Degenerative cerebellar ataxia, MS	2
Long-term outcome appears to be influenced by training intensity at home [26, 27]	Degenerative cerebellar ataxia	3
There are indications that training effects are less distinct for patients whose afferent pathways are affected [26, 27]	Degenerative cerebellar ataxia	3
Patients with mild ataxia seem to be more likely to benefit from physical therapy combined with occupational therapy than patients with more severe ataxia [38]	Degenerative cerebellar ataxia	3
Physical therapy may result in improvement of balance, gait, diadochokinesis and EDSS [2]	MS	3
Use of Johnstone Pressure Splints in addition to physical therapy does not have further benefit [2]	MS	3
Physical therapy may improve gait [3]	TBI	3
Physical therapy with the use of weighting, together with occupational therapy, can result in improvement of functional ability, fatigue and physical functioning [28]	MS	3
Treadmill training		
Treadmill training might improve gait and balance [9, 18, 53]	TBI	3
Relaxation and biofeedback therapy		
EMG biofeedback may help to reduce the amount of inappropriate coactivation of muscle groups [11]	MS, degenerative cerebellar ataxia	3
Relaxation and EMG biofeedback might decrease severity of ataxic tremor [23, 24]	TBI	3
Computer assisted training		
Adaptive robot therapy may improve coordination of the upper limbs [54]	MS	3
Supervised sports		
Climbing training might help to improve pointing movements of limbs, manual dexterity and balance [34]	Perinatal anoxic encephalopathy, TBI, stroke, metabolic encephalopathy	3
Occupational therapy		
The use of occupational therapy may diminish symptoms of depression [49]	Degenerative cerebellar ataxia	3
Occupational therapy combined with physical therapy might improve functional status [28]	MS	3
Occupational therapy combined with physical therapy might improve functional independence and gait and diminish ataxia severity and incidence of falls [38]	Degenerative cerebellar ataxia	3
Speech and language therapy		
No conclusion could be made due to insufficient data		

MS multiple sclerosis, *TBI* traumatic brain injury, *EMG* electromyogram, *ADLs* activities of daily functioning

observed improvement was related to the specific intervention, to a more generalized and non-specific element of training, to the natural disease course, or to a placebo effect such as attention. An interesting finding in one study was that only depressive symptoms improved, without further functional benefit of the applied occupational therapy [49]. For future studies, it might be valuable to examine whether the presence of depressive symptoms influences the effectiveness of such therapies, or whether possible functional improvements are driven by a reduction of depressive symptoms due to the intervention. Similarly, the influence of cognitive disturbances, which may co-exist in many diseases that underlie ataxia, should be explored. A blinded assessor was rarely used, which may have caused bias during effects measurements. Lastly, there may be a possible publication bias, as all included studies reported a positive effect.

So, what are the practical recommendations? Physical therapy and occupational therapy are recommended in cerebellar disease, based on two level 2 conclusions for

physical therapy, and the many level 3 conclusions for physical therapy and occupational therapy (Table 2). However, because of the variety of often individualized interventions and because most of the interventions were not described in full detail, it is impossible at this stage to provide a detailed guideline for such a physical therapy program. The relatively better studies suggest that the program should be intensive and adjusted to the needs and limitations of the patient, that patients should be encouraged to practice at home, and that treatment should start in earlier stages of disease, since those patients seem to be more likely to benefit. Better still is to stimulate the medical community to design and conduct randomized and controlled trials that explore the effect of allied health care interventions in neurological conditions such as cerebellar ataxia. We could learn from the work done in the field of Parkinson's disease [42].

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Conflicts of interest The authors declare that they have no conflict of interest.

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