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Review

Effect of Exercise on Physical Recovery of People with Locked-In Syndrome after Stroke: What Do We Know from the Current Evidence? A Systematic Review

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Keywords

Stroke · Exercise · Physical recovery · Locked-in syndrome

Abstract

Introduction: Locked-in syndrome (LIS) results from a brainstem lesion in the pons. Ischemic stroke is the most common etiology of LIS. People with LIS have poor mobility with serious complications due to immobilization. Benefits of exercise after stroke have been widely reported. However, little is known about what and how much exercise should be prescribed for these patients. ***Objectives:*** To explore and evaluate the effect of exercise on the physical recovery of people with LIS after stroke. ***Methods:*** We searched the following databases (last searched August 2017): EMBASE, MEDLINE, PubMed, CINAHL, AMED, PEDro, Cochrane Central Register of Controlled Trials, REHABDATA, Google Scholar, WANFANG, CNKI, and CQVIP. Handsearching of relevant journals and reference lists was also performed. The Oxford Centre for Evidence-Based Medicine was used to assess the evidence level of the included studies. ***Results:*** We identified 5 papers from 207 papers involving 35 cases; 26 cases had various degrees of improvement in physical performance after exercise; 9 cases had no change. Five types of exercises and prescriptions were adopted. Study designs and interventions were heterogeneous. All studies contained mixed rehabilitation interventions. A total of 8 different outcome measurement tools have been reported in the studies. ***Conclusion:*** Studies indicate a positive trend of effect of exercise for physical recovery of people with LIS after stroke including the improvement of muscle strength, tone, walking ability, and activity in daily living. Mixed physical exercises were used. The effects were not significant. No adverse event has

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been reported. The quality of the existing evidence is relatively low since the papers were either case series or case studies. Further studies are needed on exercise types and dosages for better prescriptions for people with LIS after stroke. This may help to extend their lives with better control of the complications and to improve their quality of life.

Introduction

Stroke is a disabling health problem which can cause loss of or limitations in physical functioning to stroke survivors. Locked-in syndrome (LIS) results from a brainstem lesion in the pons. Ischemic stroke is the most common etiology of LIS whilst the other common cause is hemorrhagic stroke [1, 2]. Bauer et al. [3] classified LIS into 3 subtypes: classic LIS, incomplete LIS, and complete LIS. Features of classic LIS are total immobility with remnants of vertical eye movement and blinking. For patients with incomplete LIS, some voluntary movements can be recovered. Patients with total LIS are totally immobilized and have no eye movement [3]. People with LIS have poor mobility with serious complications due to immobilization such as muscle atrophy, joint contracture, cardiopulmonary disorders, and urinary tract infection, of which pulmonary complications have been reported as the leading cause of death [1, 2, 4].

Benefits of exercise for stroke rehabilitation have been widely reported. Physical exercises can improve mobility, functional recovery, functional capacity, and the ability to carry out activities of daily life, and decrease the risk of cardiovascular complications after stroke [5, 6]. However, little is known about what and how much exercise should be prescribed for people with LIS after stroke.

Objectives

The primary objective is to evaluate the potential effect of exercise on physical recovery of people with LIS after stroke. The secondary objective is to explore suitable exercise prescriptions for this condition from available evidence.

Methods

We searched the following databases (last searched August 2017): EMBASE, MEDLINE, PubMed, CINAHL, AMED, PEDro, Cochrane Central Register of Controlled Trials, REHABDATA, Google Scholar, WANFANG, CNKI, and CQVIP. Handsearching of relevant journals and reference lists was also performed to identify possible evidence. Any articles written in English or Chinese were reviewed. Two reviewers independently searched and selected the literature, and two other reviewers assessed the methodological quality of the included studies with reference to the eligibility criteria. A third reviewer was consulted to resolve any uncertainty and disagreement between those two reviewers concerning the identified articles. The following terms were searched to identify related articles: stroke, CVA, cerebrovascular disease(s), physical exercise(s), physical recovery, locked-in syndrome(s). A combination of the terms was used to obtain a relatively comprehensive list of articles.

The Oxford Centre for Evidence-Based Medicine "OCEBM Levels of Evidence" 2011 version [7] was used to appraise the evidence level of the included studies. The National Institutes of Health (NIH) Quality Assessment Tool for Case Series studies [8] was used to assess the quality of the case studies included in this review.

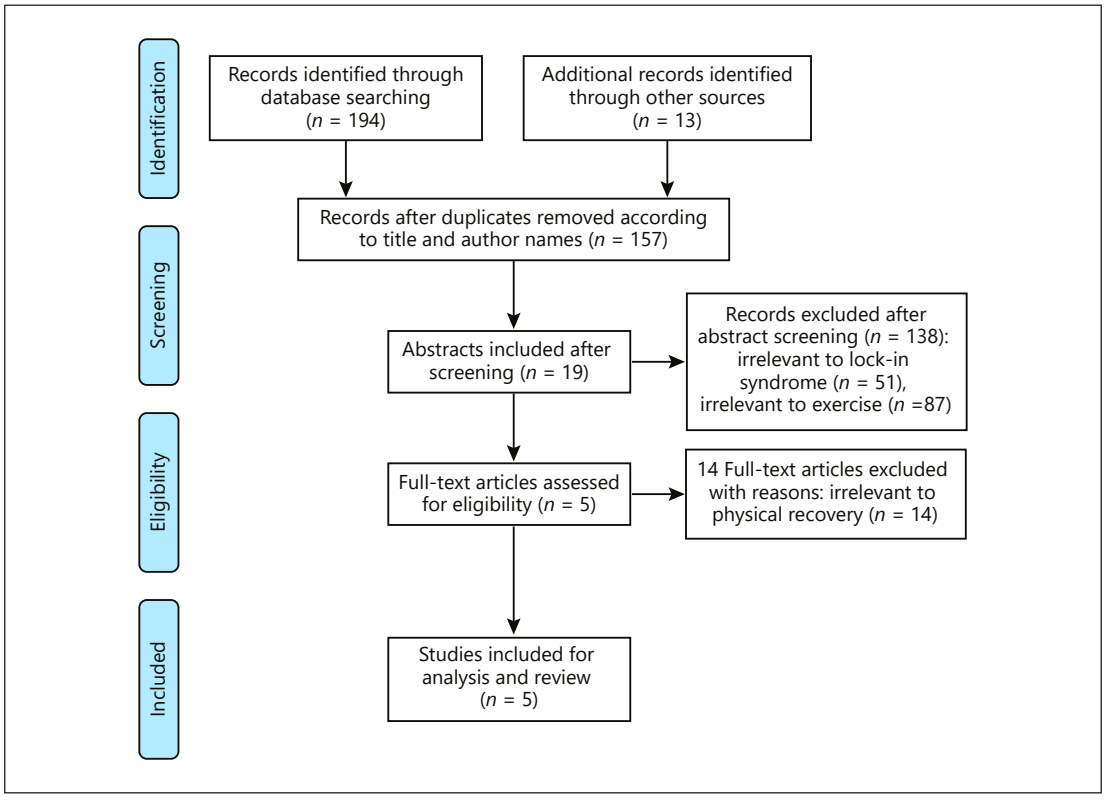

Fig. 1. Flow diagram of selection process.

Results

A total of 207 papers were identified, from which 5 papers were retrieved and retained for this review; 35 cases are involved in the included papers. All of the included studies are case studies, 4 of which are written in English and 1 in Chinese. The selection process is shown in Figure 1. A total of 22 males and 13 females were included in the studies (age range 16–72 years).

The designs and interventions of the included studies were heterogeneous. Five types of exercises and prescriptions were adopted. All studies contained mixed rehabilitation interventions. A total of 8 different outcome measurement tools have been reported in the studies including amount of body weight support (n = 1), Barthel index (n = 1), Fugl-Meyer test (n = 1), Glasgow Coma Score (n = 1), Modified Ashworth Scale (n = 1), Patterson and Grabois classification (n = 2), personal assistance (n = 1), and walking distance (n = 1). Characteristics of the studies are summarized in Table 1.

The included studies suggest a positive trend of beneficial effect of exercise on physical recovery of LIS after stroke. No adverse effect was reported. There was no report of admission to hospital or higher dependency healthcare unit or institution. Of the 35 cases, 26 cases had various degrees of improvement in physical performance after exercise, and 9 cases had no change.

According to the Oxford Centre for Evidence-Based Medicine scale, the included studies were at level 4, which is relatively low. According to the NIH quality assessment tool, 1 of the included studies was rated as fair in overall quality whilst the other 4 studies were poor in quality. Statistical methods were not well described in the studies.

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Table 1. Summary of study characteristics

Author, Year	Study design (participants)	Age range, years (gender)	Classification of LIS and etiology	Location of lesion (CT/MRI)	Exercise intervention	Other intervention	Frequency and duration	Outcome measures	Result
Hoyer et al. [9], 2009	Case series (*n* = 9)	41–72 (6 M, 3 F)	Incomplete (*n* = 9) Etiology: vascular (*n* = 9)	Pons (*n* = 7) Brainstem, caudal brainstem (*n* = 2)	Treadmill therapy	Occupational therapy, speech and swallow therapy, and psychology	1 h/day, 5 days/week	Amount of body weight support, personal assistance, and walking distance	Reduced body weight support and decreased personal assistance, increased walking distance Improved physical performance
Hummelsheim et al. [10], 1999	Case study (*n* = 1)	45 (1 M)	Incomplete (*n* = 1) Etiology: vascular (*n* = 1)	Pons (*n* = 1)	Repetitive sensori-motor training	Passive cycling, occupational therapy	60 and 70 min for each arm 5 days/week 78 weeks	Fugl-Meyer test, Modified Ashworth scale	Improved muscle strength, decreased muscle tone, improved motor function
Casanova et al. [11], 2003	Case series (*n* = 14)	16–71 (9 M, 5 F)	Complete (*n* = 3) Classic (*n* = 11) Etiology: traumatic (*n* = 3) vascular (*n* = 11)	Pons (*n* = 7) Pons, brainstem (*n* = 7)	Muscle strengthening	Respiratory, swallowing, and speech training	4 sessions/day, 6 days/week	Patterson and Grabois classification	Improved motor recovery: full in 1 case (7%), moderate in 2 cases (14%), minimal in 8 cases (57%), none in 3 cases (21%)
Kearney et al. [2], 2011	Case study (*n* = 1)	37 (1 F)	Incomplete (*n* = 1) Etiology: vascular (*n* = 1)	Pons, cerebellum (*n* = 1)	Walking exercise	OT and speech therapy	4 sessions/day, 18 weeks	Patterson and Grabois classification	"Full recovery" of motor function
Zhang et al. [12], 2009	Case series (*n* = 10)	25–64 (6 M, 4 F)	Classic (*n* = 9) Complete (*n* = 1) Etiology: traumatic (*n* = 1) vascular (*n* = 8) Hypoglycemia and ion disorder (*n* = 1)	Pons (*n* = 7) Midbrain (*n* = 1) Pons, callosum (*n* = 1)	Limb mobilization	OT, swallowing, and speech training Traditional therapy	5 sessions/day 5 days/week	Barthel index, Glasgow Coma Score	2 (20%) significant improvement, 2 (20%) improvement, 6 (60%) no change

LIS, locked-in syndrome; M male; F, female; OT, occupational therapy.

Discussion

Physical exercises may be beneficial for physical recovery of people with LIS after stroke. However, current evidence remains insufficient to draw a defined conclusion about the efficiency of exercise for physical recovery of LIS after stroke. Whilst the existing evidence is either from case series or case studies, potential bias in the evidence may have influenced the results. Different types of evidence provide valuable implications to inform practice and future research on a specific health issue. Although the quality of the relevant existing evidence is relatively low, useful information may still be extracted from the studies. Systematic appraisal and review of case studies remain valuable for suggesting clinically meaningful information for patient-centered care. This is particularly important for rare diseases and special health conditions, of which high-level evidence may be insufficient whilst they may be less well studied.

In view of evidence-based practice, clinicians should integrate knowledge from scientific research, their professional judgement from related clinical expertise and experiences, and individual patients' actual needs to design safe and specific exercise programs for these patients. As papers written in languages other than English and Chinese were not included, publication and language bias may exist in this review. Research is still needed to unlock the secret of exercises for LIS and to determine specific guidelines to promote the physical recovery of stroke survivors with LIS. Studies are also needed to explore the role of emerging assistive technologies in enhancing the delivery of personalized exercise interventions for stroke survivors with LIS.

Conclusion

The reviewed studies indicate a positive trend of effect of exercise on physical recovery of people with LIS after stroke including the improvement of muscle strength, tone, walking ability, and activity in daily living. No adverse event was reported. Study designs and current interventions were heterogeneous. A wide range of outcome measures was used. The effects reported in the studies were not significant while the methodological quality of the existing evidence was relatively low. Well-designed studies are needed to determine the effect of exercise and suitable exercise prescriptions for the rehabilitation needs of people with LIS after stroke.

Disclosure Statement

The authors have no conflicts of interest to declare in relation to this article.

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