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Research and theory

Economic evidence on integrated care for stroke patients; a systematic review

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Abstract

Introduction: Given the high incidence of stroke worldwide and the large costs associated with the use of health care resources, it is important to define cost-effective and evidence-based services for stroke rehabilitation. The objective of this review was to assess the evidence on the relative cost or cost-effectiveness of all integrated care arrangements for stroke patients compared to usual care. Integrated care was defined as a multidisciplinary tool to improve the quality and efficiency of evidence-based care and is used as a communication tool between professionals to manage and standardize the outcome-orientated care.

Methods: A systematic literature review of cost analyses and economic evaluations was performed. Study characteristics, study quality and results were summarized.

Results: Fifteen studies met the inclusion criteria; six on early-supported discharge services, four on home-based rehabilitation, two on stroke units and three on stroke services. The follow-up per patient was generally short; one year or less. The comparators and the scope of included costs varied between studies.

Conclusions: Six out of six studies provided evidence that the costs of early-supported discharge are less than for conventional care, at similar health outcomes. Home-based rehabilitation is unlikely to lead to cost-savings, but achieves better health outcomes. Care in stroke units is more expensive than conventional care, but leads to improved health outcomes. The cost-effectiveness studies on integrated stroke services suggest that they can reduce costs. For future research we recommend to focus on the moderate and severely affected patients, include stroke severity as variable, adopt a societal costing perspective and include long-term costs and effects.

Keywords

stroke, integrated care, rehabilitation, costs, economics, review

Introduction

Annually, 15 million people worldwide suffer from a stroke. Of these, five million die and another five million become permanently disabled [1]. Many persons who have survived a stroke experience problems in one or more health-related domains, such as physical, cognitive, behavioural and emotional well-being. With an

aging population and improved treatment options, this number is only expected to increase. In many Western countries, stroke is the biggest single cause of disability [1]. It should not come as a surprise that the economic burden of stroke is very large. Apart from the costs to the health system and rehabilitation services, there are also the production losses to society rising from premature deaths and disability, and costs to the individual and families who have to take time off from work to provide informal care. The extent of the problem, combined with limited health care budgets emphasizes the need for an evidence-based and cost-effective health care delivery in stroke care.

Theory

Stroke care is complex and covers a whole spectrum of care including acute care, rehabilitation and long-term care with both in-hospital, outpatient and community-based care. Because of all these different types of care; stroke care is per definition multidisciplinary and the integration of care is of particular concern [2]. Ideally, optimal stroke care integrates all relevant care providers; hospitals, nursing homes, rehabilitation centres, general practitioners and home care providers who work together to provide multidisciplinary, coordinated care through organized patient transfers and protocols. In the last years, there has been a call to improve health care delivery to patients in terms of greater consistency, effectiveness, care continuity and improved collaboration [3].

In light of these developments, various ways of organizing and delivering care for stroke patients have been implemented. In this review, we focus on all stroke care *beyond* emergency (pre-)hospital care. Two Cochrane reviews concluded that more organized inpatient care for stroke patients was consistently found to yield better health effects [4, 5]. However, it remains unclear which way of delivering and coordination of comprehensive and multidisciplinary post-emergency care along the disease continuum and across the different health care systems is the most cost-effective.

Previous reviews into the cost-effectiveness of integrated stroke care have looked at only one type of integrated care arrangement, or looked only at in-hospital costs [4, 6, 7]. When studying integrated care interventions for stroke, it is important to take a wider perspective and include other costs, such as rehabilitation costs, informal caregiver costs and indirect costs, to be able to detect a possible cost shift. An intervention that is cost-effective for the hospital might put a larger financial burden on the nursing home, rehabilitation service or on the informal caregivers, the family and the society at large.

A comprehensive review collecting the available evidence on the cost-effectiveness of different integrated stroke care arrangements and their health outcomes can provide more insight into this issue. This review aims to provide an overview of the economic evidence of the currently existing types of post-emergency integrated care arrangements for stroke patients.

Methods

Study selection criteria

The current review is restricted to empirical studies that provide quantitative data, thus excluding qualitative studies, reviews and case reports. Only studies of stroke populations were included in which integrated care was studied, and where an economic evaluation was reported (either costs or resource use with mentioning of unit prices). The quality of the economic studies was evaluated by the researchers and studies of low quality were excluded from the final analysis (see Appendix).

Integrated care arrangement: To ensure we included all currently existing integrated care arrangements, we adopted the broad definition of integrated care of the European Pathway Association; 'Integrated care is a multidisciplinary tool to improve the quality and efficiency of evidence based care which is used as a communication tool between professionals to manage and standardize the outcome orientated care.' [3].

Economic evaluation: Primary studies that were either a full economic evaluation or a cost analysis were included, following the classification of economic studies of Drummond [8]. Studies that provided only a cost-outcome description, cost description or modelled costs were excluded. The review includes economic studies along the full spectrum of services—acute care, rehabilitation and long-term support.

Literature search strategy

The databases of MEDLINE and EMBASE were searched in parallel for relevant English articles published up to July 11th, 2011. The search terms used for 'economic evaluation' were: economic analysis, economic evaluation, cost-benefit, cost-effectiveness, costs, health resources, resource utilization and utilization of health care. The search terms for 'integrated stroke care' were stroke service, stroke unit, integrated stroke care, organized stroke care, stroke rehabilitation, integrated care pathway, clinical pathway, critical pathway and disease management. Only articles that included both the economic information and the integrated stroke care element were examined further.

Additional studies were identified through reference lists and overview articles.

The search strategy produced a total of 415 articles. After removing 150 duplicates, a further 179 articles were removed after screening titles. Screening abstracts and full-text led to the removal of 68 more articles (including eight articles from which we failed to obtain the complete article [9–16]). Four studies were considered to be of low quality, and were therefore excluded from the final analysis [17–20] (see Appendix). This left a total of 15 studies for inclusion in the final review.

Data extraction and synthesis

Two researchers (JT and AS) independently selected studies; any disagreement was resolved through consensus. One reviewer (JT) extracted information from the selected studies.

Given the heterogeneity of the included studies, a qualitative approach summarizing the characteristics and results of the selected studies was used for data synthesis. The results of the studies are not pooled quantitatively, given the variation in study characteristics and methodology.

From the articles, information was extracted on study type, intervention details, control details, number of subjects, length of follow-up, type of facilities included in the cost analysis, type of economic study, costing perspective, mean cost per patient, type of cost included, how costs were determined and whether the authors had conducted sensitivity analysis. The direction and magnitude of total costs per patient in the intervention relative to the comparator were tabulated.

Several studies that reported a cost analysis had published health outcomes of the trial elsewhere. To provide a comprehensive overview, the health outcomes of these studies were also included in the final results table in this review.

The level of evidence concerning the cost-effectiveness for each service type was determined subjectively by weighing the number of studies, the consistency of cost trends, the 'robustness' of the results and the methodological quality of the study.

Intervention categories

The interventions reported in the fifteen studies were grouped into four categories, which were deduced from the material; 1) early supported discharge (n=6), 2) home-based rehabilitation (n=4), 3) stroke unit care (n=2), and 4) stroke service from acute to chronic phase (n=3). The intervention categories are associated with different parts of the rehabilitation process, although there can be some overlap between interventions. For example, some interventions that focus on early-supported discharge also include home-based rehabilitation, or rehabilitation in a day-hospital. The economic evidence will be presented separately for each intervention category.

A short description of characteristics of each intervention category is provided here. In early-supported discharge, a multidisciplinary team facilitates discharge in order to reduce the acute hospital stay [21, 22]. In home-based rehabilitation, coordinated multidisciplinary rehabilitation takes place in the house of the patient [23]. In a stroke unit, care is organized by using clinical pathways for diagnosis, treatment, prevention of complications and rehabilitation specifically for stroke care. In addition, multidisciplinary teams coordinate care, rehabilitation therapy and patient education aimed at reaching predefined goals before the patient is discharged [24]. A stroke service was defined by one study as 'a network of service providers, working together in an organized way to provide adequate services in all stages of the follow-up of stroke patients [25]. It includes a hospital stroke unit and at least one other service provider.

Results

Study characteristics

Ten out of fifteen studies were set in Europe (of which five were in the UK) [22, 25–33], two in Australia [34, 35] and Canada [36, 37], and one in Hong Kong [38] (Table 1). The time horizon of the studies was generally short; most of the studies followed the subjects for a year (n=9), and the rest for a shorter period (n=6). Most (n=12) of the included studies were randomized controlled trials, and three were non-randomized. The study size ranged from 83 to 598 subjects.

Even though 11 out of the 15 included studies were published after 2000, for all but one study [37] the data were collected from trials performed before 2000.

The intervention and the comparators are described in detail in Table 2 and the economic characteristics of the included studies in Table 3. Six studies were classified as 'cost analysis', seven studies as 'cost-effectiveness study' and two as 'cost minimization analysis' (although some were labelled differently by their authors). The scope of costs included varied, with only four studies [31, 32, 34, 37] using the preferred societal perspective, and the rest (n=11) adopting a health care perspective, including only costs related to the health care system.

Table 1. Summary of study characteristics.

Study characteristics	No. of studies
Intervention category	
Early supported discharge	6
Home-based rehabilitation	4
Stroke unit care	2
Stroke service	3
Study setting—continent	
Europe	10
Other	5
Follow-up	
1 year	9
6 months	6
Type of study	
RCT	12
Non-randomized trial	3
Size of study population	
N<100	3
n=100–300	6
n>300	6
Costing perspective	
Health care perspective	11
Societal perspective	4
Date of publication	
Before 2000	4
After 2000	11
Data collection	
Before 2000	14
After 2000	1

Economic results

In this section, an overview is presented of the economic results per intervention category (Table 4).

Early-supported discharge

In many places, early discharge from the hospital is widely practiced and encouraged. However, the degree to which discharge is facilitated or aftercare is arranged, differs greatly between settings [39]. The six studies reporting on the costs of early-supported discharge investigated slightly different versions of early supported discharge services. The interventions have in common that all included at least a multidisciplinary team, and they all aimed to reduce the length of acute-hospital stay.

For all six studies, the intervention did not result in worse health outcomes when compared to the comparator. Early-supported discharge combined with home rehabilitation even resulted in a significantly higher SF-36 score in the study of Teng et al [36].

Six out of six studies showed that early-supported discharge resulted in lower costs, in the range of a 4–30% cost reduction compared to usual or conventional care, but only in the study of Teng et al. [36], this difference was significant. Anderson et al. [34] report that although the cost reduction of 20% was not significant, sensitivity analysis found consistently lower costs for the intervention. Both Anderson [34] and McNamee [27] reported that the costs of the remaining care at home significantly correlated with a patient's level of disability; those with mild disability scores had significantly lower costs compared to those with moderate disability scores, even after adjustment for age, co-morbidity, and presence or absence of a caregiver.

Beech et al. [26] considered it unlikely that early-supported discharge leads to financial savings, but due to a shorter length of stay it might release capacity for an expansion in caseload, which can also be a desirable outcome. In support of this hypothesis, both McNamee et al. [27] and Van Koch et al. [28] reported a significant reduction in the length-of-stay, which most likely explains the reported reduction in costs.

Home-based rehabilitation

The four studies that compared home-based rehabilitation were set in the UK [29, 30], Canada [37] and Sweden [33]. Gladman et al. [29] and Andersson et al. [33] both compare home-based rehabilitation to hospital-based rehabilitation, Roderick et al. [30] compared the home-based rehabilitation to care in a geriatric day-hospital, and Markle-Reid et al. [37] compared the delivery of home-care by a specialized team to usual home care. All the home-based care interventions have in common that the care at home was delivered by a multidisciplinary team.

Three out of four studies [29, 30, 37] reported non-significant higher costs for the intervention, accompanied by a significant improvement in the Barthel Index for Roderick et al. [30] and a clinically important improvement in the SF-36 score in the intervention group reported by Markle-Reid [37]. In the study of Gladman et al. [29] younger patients seemed to benefit more from home-care and older patients from hospital-based care.

In contrast to the three other studies, Andersson et al. [33] reported similar costs for home-based rehabilitation compared to hospital rehabilitation. The authors of this study suggested that home-based rehabilitation might in fact turn out to be costs-saving since most patients who should receive home-based rehabilitation had to wait longer in the expensive acute care beds of the hospital compared to the hospital-based group, because the necessary adjustments had not taken place in their houses. The authors suggest that a more smooth transition will render home-based rehabilitation cost-effective.

Home-based rehabilitation is often presented as a less expensive alternative for inpatient rehabilitation, as expensive traditional hospital care is substituted by less expensive care in the patient's home [33, 37]. The results presented above, however, suggest the opposite. Possible explanations for the lower costs of

Table 2. Study characteristics, intervention and control details

Author; year (country)	Intervention	Control(s)	Study design - Type - Follow up period	Intervention and control details
1) Early supported discharge (ESD)				
Anderson; 2000 (Australia) [34]	ESD and home-based rehabilitation (n=42)	Usual care (n=44)	RCT (n= 86) - 6 months	**Intervention:** ESD with rehabilitation at home provided by multidisciplinary team, including a coordinator **Usual care:** Conventional in-hospital rehabilitation and use of community services following the acute care period in the hospital
Beech; 1999 (UK) [26]	ESD with community-based therapy (n=167)	Usual care (n=164)	RCT, (n=331) - 1 year	**Intervention:** ESD with a planned program of care up to 3 months with a multidisciplinary ESD team consisting of a physiotherapist and therapy aide, an occupational therapist, a speech and a language therapist. **Usual care:** hospital-based stroke clinic, a geriatric day hospital and generic therapy services
Hui; 1995 (Hong Kong) [38]	ESD with geriatric team using day hospital facility	Usual care	RCT (n=120) - 6 months	**Intervention:** ESD followed by rehabilitation in a geriatric day hospital with a multidisciplinary geriatric team encompassing medical, nursing, remedial and psychological support, including input from social services and chiropodists. **Usual care:** Inpatient care on a stroke ward under a neurology team and conventional inpatient rehabilitation
McNamee; 1998 (UK) [27]	ESD (n=46)	Usual care (n=46)	RCT (n=92) - 6 months	**Intervention:** ESD with an interdisciplinary team with a service coordinator, physiotherapist, occupational therapist, speech therapist and social worker. Input from a district nurse and occupational therapy technician was obtained if required. The team organized discharge and community rehabilitation arrangements during hospitalization **Usual care:** Not reported
Teng; 2002 (Canada) [36]	ESD with home rehabilitation (n=58)	Usual care (n=56)	RCT (n=114) - 3 months	**Intervention:** ESD with a 4-week tailor-made home program of rehabilitation and nursing services by a multidisciplinary team offering nursing, physical therapy, occupational therapy, speech therapy and dietary consultation. **Usual care:** Current practices for discharge planning and referral for follow-up services such as extended acute-care hospital stay, inpatient rehabilitation, outpatient care, private care and home care by a local community health centre.
Von Koch; 2000 (Sweden) [28]	ESD and continued rehabilitation at home (n=42)	Usual care (n=41)	RCT (n=83) - 1 year	**Intervention:** ESD with tailor-made rehabilitation at home by an outreach team of occupational, physical and speech-and-language therapist, for up to 4 months **Usual care:** Rehabilitation in the stroke unit until discharge and if required in the geriatrics or rehabilitation departments as inpatients or in day care – no standardized program.
2) Home-based rehabilitation				
Andersson; 2002 (Sweden) [33]	Home-based rehabilitation (n=53)	Hospital-based rehabilitation (n=68)	Non-randomized trial (n=123) - 1 year	**Intervention:** After the acute care period in the hospital, the patient received treatment from a physiotherapist, occupational therapist, nurse, hospital social worker and speech therapist at home **Hospital-based:** After the acute care period in the hospital, patients were transferred to a rehabilitation ward in another hospital. No special arrangements were made for the studied population.

Table 2 Continued

Author; year (country)	Intervention	Control(s)	Study design - Type - Follow up period	Intervention and control details
Gladman; 1994 (UK) [29]	Home-based rehabilitation (n=162)	Hospital-based rehabilitation (n=165)	RCT (n=327) - 14 months	**Intervention:** Rehabilitation at home provided by a rehabilitation team, including a physiotherapist and occupational therapist who assessed patients at home and arranged or provided appropriate therapy or organized other necessary care for up to six months, after which routine services were used. **Hospital based rehabilitation:** Rehabilitation was provided via routine outpatient rehabilitation departments or a day-hospital
Markle-Reid; 2011 (Canada) [37]	Home-based rehabilitation from a specialized team (n=52)	Usual home care (n=49)	RCT (n=101) - 1 year	**Intervention:** Standard home care services plus home visitation by a team including a care coordinator, nurse, physiotherapist, occupational and speech therapist, dietician, social worker and personal support worker. The team used an evidence-based approach using a screening protocol, stroke education, caregiver support, monthly case conferencing and development of community reintegration plan. **Usual care:** Standard home care services including follow-up by a care coordinator who arranged home care services.
Roderick; 2001 (UK) [30]	Domiciliary rehabilitation service (n=66)	Geriatric day-hospital care (n=74)	RCT (n=140) - 6 months	**Intervention:** Domiciliary stroke team with a senior physiotherapist, senior occupational therapist and a consultant geriatrician who met daily and used a goal-setting approach. **Day hospital:** Care was coordinated by multi-disciplinary teams who gave therapy in both individual and group sessions.
3) Stroke unit care				
Moodie; 2006 (Australia) [35]	Stroke care unit (n=102)	Conventional in-hospital care (n=84) OR stroke team (n=209)	Prospective cohort study (n=395) - 6 months	**Stroke unit:** A designated ward area with a dedicated multidisciplinary stroke team **Stroke team:** A dedicated, hospital-based multidisciplinary team, which reviews stroke patients located in different wards. **Conventional care:** General medical ward care with no dedicated stroke service or health professional team
Patel; 2004 (UK) [32]	Stroke unit (n=152)	Stroke team (n=152) OR domiciliary care (n=153)	RCT (n=457) - 1 year	Patients were randomized within 72 hours after stroke onset into one of the three intervention groups: **Stroke unit:** 24 hours care by a specialist multidisciplinary team based on clear guidelines for acute care, prevention of complications, rehabilitation and secondary prevention, including discharge planning. After discharge treatment was provided by community services. **Stroke team:** Management on general wards with specialist stroke team support who assessed and advised ward-based nurses and staff on acute care, secondary prevention and rehabilitation. After discharge treatment was provided by community services. **Domiciliary care:** Management at home under supervision of a specialist team, GP and stroke physician; investigations performed on an outpatient basis; therapy handled by specialist staff; district nursing support was available and personal care from social services for max. 3 months.

Table 2 Continued

Author; year (country)	Intervention	Control(s)	Study design - Type - Follow up period	Intervention and control details
4) Stroke service				
Claesson; 2000 (Sweden) [31]	Stroke unit linked to continued care in geriatric stroke units (n=166)	Usual care (general medical ward) (n=83)	RCT, (n=249) -1 year	**Stroke service:** A non-intensive acute stroke unit integrated with continued care in geriatric stroke units after discharge from the hospital, when continued care was needed. The two units worked according to identical principles, which had been agreed on. The stroke unit included a multidisciplinary stroke unit team, also involved in discharge planning. **Usual care:** Conventional acute medical care in a general ward, not in the framework of a structured stroke unit care approach. There was no standardized treatment or extra resources available for management of stroke patients.
Fjaertoft; 2005 (Norway) [22]	Extended stroke unit service (n=160)	Ordinary stroke unit service (n=160)	RCT (n=320) - 1 year	**Intervention:** An extended stroke unit service with early supported discharge, cooperation with the primary healthcare system and more emphasis on rehabilitation at home. A mobile team coordinated the stroke service and established an individual program and support system, and consisted of a physiotherapist, occupational therapist, a nurse and a physician **Control:** Conventional stroke unit care with follow-up organized by rehabilitation clinics and/or the primary health care system.
Van Exel; 2005 (Netherlands) [25]	Stroke service in three centers; Delft, (n=151) Haarlem (n=111) and Nijmegen (n=149)	Usual stroke care (n=187)	Non-randomized cluster trial (n=598) - 6 months	**Delft:** Stroke service including a hospital with a stroke unit, a nursing home, specialized nurses in home care and transmural stroke nurses responsible for patient transfers **Nijmegen:** Stroke service including two hospitals, various nursing homes, post-acute care at home and specialized stroke care training for nurses **Haarlem:** Stroke service including three hospitals, a nursing home and specialized home rehabilitation

RCT, Randomized controlled trial; ESD, Early supported discharge; LOS, length of stay; GP, general practitioner; UK, United Kingdom.

Table 3. Characteristics of the economic analysis.

Author; year	Type of economic study	Costing perspective	Costs included	Sensitivity analysis	How were costs determined?
1. Early supported discharge					
Anderson; 2000 [34]	Cost minimization	Societal	Direct and indirect costs of health care, rehabilitation, patients and ICG	Yes	• Community costs and caregiver costs (personal expenses) obtained via questionnaire • Hospital costs via hospital records and rehabilitation costs via staff records • No production losses included
Beech; 1999 [26]	Cost analysis	Healthcare	Patient utilization of health and social services, hospital costs	Yes	• In-hospital and ESD team costs were determined via data from provider departments and local finance departments. All costs included over-head costs
Hui; 1995 [38]	Cost-effectiveness	Healthcare	Use of acute and rehabilitation beds, and number of days of GDH attendance and outpatient clinic visits	No	• Fixed prices derived from local data were used as unit costs • Costs intervention group = total inpatient stay + GDH attendance + out-patient clinic attendance + hospital readmission • Costs comparison group = total inpatient stay + outpatient clinic attend-ance + hospital readmission
McNamee; 1998 [27]	Cost analysis	Healthcare	Health services, social services, rehabilitation costs, travel time per visit	Yes	• In-hospital costs; weighted average (based on relative physical depen-dency) specialty costs per bed day • Rehabilitation costs; local staff unit costs per hour • Other services; unit costs per visit
Teng; 2002 [36]	Cost analysis	Healthcare	Acute care costs, home intervention costs, usual care costs, readmission costs, physician	Yes	• Unit costs for services were calculated as average costs, including overhead costs and allowance for opportunity cost of building and land • Physician, emergency room attendance and readmissions were obtained via fee-for-service billings from a local finance department
Van Koch; 2000 [28]	Cost-effectiveness	Healthcare	Inpatient hospital care, outpatient health care, use of health-related services, and cost of health care	No	• Data on hospital stay and number of outpatient or home rehabilitation visits was collected from a local council register. • Data on community-based services and informal care was obtained via a patient or spouse interview. • Average cost per type of service was obtained from the Stockholm County Council statistics
2. Home-based rehabilitation					
Andersson; 2002 [33]	Cost analysis	Healthcare	Acute care, rehabilitation cost in-hospital and home-based, home-help services and nursing home costs	No	• Costs for acute care were fixed costs per day • Costs for in-hospital rehabilitation were fixt costs per day • Costs for the home-based rehabilitation was based on time spent with the patient, and included overhead • Prices for home-help were market prices set by procurement procedure
Gladman; 1994 [29]	Cost analysis	Healthcare	Domiciliary team, day hospital and outpatient attendances	No	• Information about indirect costs was gathered but not incorporated into the cost analysis; no significant differences between the groups • Domiciliary costs; gross employment costs of the therapists plus vehicle running costs. Hospital costs; internal accounts provided day-hospital and out-patient rehabilitation costs
Markle-Reid; 2011 [37]	Cost-effectiveness	Societal	Primary care, emergency department, hospital days, health and social	No	• Quantities of use of health services were determined using the Health and Social Services Utilization Inventory • Price data from 2006 were used

Table 3 Continued

Author; year	Type of economic study	Costing perspective	Costs included	Sensitivity analysis	How were costs determined?
			professional costs, medications, lab services, indirect costs including pension, worker's compensation, employment insurance and private insurance		
Roderick; 2001 [30]	Cost-effectiveness	Healthcare	Health service and social service costs including transport costs	Yes	• Service use: determined via hospital records, nurse records, GP records, social services records and a special designed domiciliary service form. • Transport costs: ambulance and therapists' travel
3. Stroke unit care					
Moodie; 2006 [35]	Cost-effectiveness	Healthcare	Health sector resource use including pre- and post-hospital resource use	Yes	• Micro-costing approach using individual patient resource-use data • Resource use attributable to the index stroke was measured, including pre-hospital referral and transport costs obtained via self-report or hospital medical records. Acute transmission costs were obtained via the hospital finance department, including indirect costs. Post-hospital resource use was recorded in a diary
Patel; 2004 [32]	Cost-effectiveness and cost-utility	Societal	Health care, social services and informal care	Yes	• Hospital resource use, therapy input were recorded on an ongoing basis, data on public sector services and informal care were obtained via a patient interview. • Informal costs were estimated 1) via the UK minimum wage rate and 2) based on the unit cost of a social services home help worker
4. Stroke service					
Claesson; 2000 [31]	Cost minimization	Societal	Hospitalization costs, institutionalized living, outpatient care, different kinds of support, ICG	No	• Informal care costs were calculated with unit cost per hour; 35% of gross wage rate • Hospital data from hospital records • Outpatient service use, use of additional support and number of hours of informal care were assessed via a patient-questionnaire
Fjaertoft; 2005 [22]	Cost analysis	Healthcare	Use of health services and hospital expenses	Yes	• Prospective recording of health service use • Use of health services; costs measured as service costs. Hospital expenses measured as costs per inpatient day, adjusted for DRG
Van Exel; 2005 [25]	Cost-effectiveness	Healthcare	Care utilization in hospitals, nursing homes and rehabilitation centres, outpatient care, paramedical care	Yes	• Actual costs of resource use were determined using individual health care utilization and unit costs for health resources. • In-hospital data obtained via medical patient files, outpatient care data via patient interviews

ICG=Informal care giver; DRG=Diagnosis-related group; GDH=Geriatric day-hospital; ESD=Early supported discharge.

Tabel 4. Comparison of costs and health outcomes reported by intervention category.

Author	Cost per patient					Cost reduction for intervention, %		Health Outcome
	Valuta	Intervention		Comparison				
1. Early supported discharge								
Anderson et al.	AUD$	8040		10,054			**20**	NS
Beech et al.	£	6800		7432			**8**	NS
Hui et al.	HK$	58,168		51,809			**12**	NS
McNamee et al.	£	7155		7480			**4**	NS
Teng et al.	C$	7784		11,065			**30****	Better SF-36 score [21]
Van Koch et al.	SEK	71,959		91,453			**21**	NS
2. Home-based rehabilitation								
Andersson et al.	SEK	190,500		194,700			**2**	NS
Gladman et al.	£	408		320			**-27**	NS
Markle-Reid et al.	C$	32,791		30,937			**-6**	NS, clinically important change in SF-36
Roderick et al.	£	4240		3,574			**-18**	Increased change in Barthel Index
3. Stroke unit care								
Moodie et al.	AUD$	15,383		CC: ST:	12,251 15,903	SCU: ST:	**-26** **-30***	Better adherence to process indicators[a] and less complications[a]
Patel et al.	£	16,574		DC: ST:	10,296 12,512	SCU:	**-60****	Fewer deaths, less institutionalization after 1 year and more QALYs gained in the SCU. ST had the worst health outcomes
4. Stroke service								
Claesson et al.	$	25,373		28,507			**11**	NS
Fjaertoft et al.	€	18,937		21,824			**13**	Better Rankin Scale score, Barthel index [41], ADL index and QoL [44] after 26 weeks
Van Exel et al.	€ € €	Delft: Haarlem: Nijmegen:	13,160 16,790 20,230	13,810		Delft: Haarlem: Nijmegen:	**5** **-22** **-46****	Delft: better HRQoL Haarlem: worst HRQoL Nijmegen: NS difference

NR, Not reported; NS, Not significant; QoL, Quality of life; ADL, activities of daily living; HRQoL, Health-related quality of life; SCU, stroke care unit; ST, stroke team; DC, domiciliary care; CC, conventional care.
Valuta: AUD$, Australian dollar; HK$, Hong Kong dollar; C$, Canadian dollar; SEK, Swedish krona; $, American dollar; £, British pound; €, Euro.
*significant at p<0.05; **significant at p<0.01.
[a]Stroke unit care versus conventional care.

inpatient care are a smaller number of staff involved, and the gaining of scale economies [29]. Such advantages were not available to the home-based rehabilitation investigated in these studies.

Roderick et al. [30] report that even though health care costs were lower for the home-based rehabilitation group, this was offset by higher social service costs. Andersson et al. [33] reported a similar cost-shift from health care providers to social welfare providers in the home-based rehabilitation group. A narrow costing perspective, looking only at costs for the health care providers may therefore paint a misleading positive financial picture.

The higher costs for the specialized team approach for home-based care in the study by Markle-Reid et al. [40] can be explained more straightforward; the team-approach had higher per-person costs of health service use compared to conventional home-care. The intervention did not pay for itself by reducing the use of expensive health care resources as the authors had expected.

From these four studies it seems likely that home-based rehabilitation after discharge will be cost-neutral from a societal perspective, and better quality of care could be achieved at similar costs.

Stroke unit care

The two studies evaluating stroke units set in Australia [35] and the UK [32]. In both of the studies, stroke units were compared to other types of interventions that could be used during the post-emergency phase of stroke care. Moodie et al. [35] compared three ways of providing in-hospital stroke care; 1) stroke unit care, 2) conventional in-hospital care, and 3) stroke care provided by an in-hospital stroke team. Patel et al. [32] also compared three interventions in the post-emergency phase; 1) stroke unit care, 2) general ward with a stroke team, and 3) stroke management at home. Both studies calculated incremental cost-effectiveness ratios (ICER) for all interventions.

In the study by Moodie et al. [35]; the costs of stroke unit care were 26% more than conventional in-hospital care, which was borderline significant (p=0.08). However, the health outcomes of stroke unit care were better than for conventional care. A more thorough adherence to process indicators and a decreased complication rate were found [35]. The incremental cost-effectiveness ratio (ICER) of stroke unit care over conventional care was AUS$9867 per patient achieving thorough adherence to clinical processes and AUS$16,372 per patient with severe complications avoided. The authors therefore concluded that dedicated stroke unit care was cost-effective. An in-hospital stroke team was the most costly intervention, and did not yield additional health gain. The study however only included patients with typically moderate stroke; and the results might not be applicable to the entire stroke population [35].

In line with the findings of Moodie et al. [35] stroke unit care was the most expensive of the three interventions in the study by Patel et al. [32], but also achieved better health outcomes. In-hospital stroke team care was dominated by stroke management at home, which was both more effective and less costly. Compared to stroke management at home, the ICER of stroke unit care was £682 of avoiding 1% of death and institutionalizations, or £89 132 per quality-adjusted life year (QALY) gained. These results should be interpreted with caution; Patel only included patients with a moderately severe stroke, who could be supported at home.

The two studies reported here yielded very similar results, and provided moderate evidence that at least for in-hospital care and for patients with a moderately severe stroke, care in stroke units yields better health outcomes. These improved outcomes come at a higher cost compared to stroke management at home, conventional in-hospital care or in-hospital stroke teams.

Stroke service

The stroke services in the three studies that we included differed in their degree of service integration. The study by Van Exel [25] for example studied three complete stroke services, while the stroke service of Claesson [31] mainly included a hospital and geriatric care component, meant for older patients. Fjaertoft [22] focused on improved transitions between acute care, early-supported discharge from a stroke unit and the primary care services with home care.

The stroke service described by Claesson et al. [31] only included patients older than 70 years, who had not been in a nursing home at the time of stroke. Their stroke service resulted in a non-significant cost reduction of 11% after the first year, with similar health outcome as the care as usual group. This study found a large variation in costs per patient, which was related to stroke severity at onset. Due to sample size restrictions it was not analyzed whether the stroke service was more cost-effective for certain patient groups based on stroke severity.

Fjaertoft et al. [22] observed a similar cost reduction of 13%. The patients who received care via the integrated stroke service also had a significantly higher Rankin scale (65 vs. 52% independence; p=0.02), Barthel index (60 vs. 50% independent in ADL; p=0.05) (41) and higher Quality-of-Life (mean score 78.9 vs. 75.2; p=0.05) [22].

Van Exel [25] evaluated three different stroke services in the Netherlands. The services all had different

organizational characteristics and included different elements. The stroke service in Delft was consistently characterized as the most organized, and with the most clear-cut patient trajectory. Compared to usual care, the stroke service in Delft was also the only one of the three stroke services that showed a minimal reduction (5%) in costs together with a better health outcome. The stroke service in Nijmegen cost 50% more, at similar health outcomes. The stroke service in Haarlem was 22% more expensive, with worse health outcomes.

Two out of three studies reported a cost reduction, against similar or better health effects. The large differences in the organization of the stroke services in the three studies can explain the different results in both costs and outcomes.

Discussion and conclusions

This review investigated economic evidence for four types of integrated care arrangements. In conclusion, six out of six studies on early supported discharge reported reduced costs with similar (n=5) or better (n=1) health outcomes. Apart from possible cost-savings, early supported discharge has the potential to free-up capacity for acute care. Home-based rehabilitation after stroke is most likely cost-neutral from a societal perspective, and can lead to improved quality of life. Two studies provided evidence that the use of stroke units yields better health outcomes, but at a higher cost compared to conventional in-hospital care or mobile stroke teams. The three studies that reported on integrated stroke services studied services that were substantially different from each other. Nonetheless, the trend in the results suggests that service integration can indeed be cost saving for stroke care.

Our findings differ at some points from an earlier review of economic evidence on stroke rehabilitation [42]. That review concluded that the costs of care in a stroke unit were comparable to care in another hospital ward, while we found higher costs [32, 35]. For home-based rehabilitation, the authors of the review concluded that studies showed conflicting results. For ESD however, their findings were in line with ours. We recognize that for integrated care arrangements, it might be true that '*the devil is in the details*', meaning that two seemingly similar programs can have very different effects because of differences in context (e.g., size of the hospital), or implementation (e.g., culture differences). An evaluation of integrated care should therefore take those variables into account. We attempted to tackle this issue by providing detailed information about the precise intervention and the setting, for as much as it was described in the articles.

Implications for future research

More evidence is needed to determine if and which integrated care arrangements are cost-effective for stroke patients. For future research, we recommend to take the following key points into account:

Include stroke severity as variable

From both a clinical and rehabilitation perspective, it makes sense to divide the stroke population into three groups; those who are mildly, moderately and severely affected. Division in these groups allows for the optimal use of resources [43]. For those who are mildly affected, early supported discharge and rehabilitation at home will probably be sufficient for recovery. For those severely affected, intensive rehabilitation programs in a rehabilitation centre might not have sufficient effect, and this group might be best off in a nursing home. The middle group, those who are moderately affected are the most interesting from a cost-effectiveness perspective. Specifically for this group of patients, it is important to find out which type of rehabilitation care (e.g., at home, in a rehabilitation centre or in a nursing home) yields the best health effects at the lowest costs. Since stroke severity is such an important variable in the outcome and effectiveness of stroke interventions, we therefore recommend that 1) future research should take stroke severity into account when researching cost-effectiveness and 2) cost-effectiveness studies for stroke interventions should focus on the patient groups with moderate and severe stroke.

Adopt a societal costing perspective

A variety of costing perspectives were used in the included studies. Besides creating a problem for comparison, this also poses a problem for the reliability of the findings; omission of certain costs can greatly influence the relative cost-effectiveness. When a health care perspective is used for example, the indirect costs and costs to informal caregivers are not included. A home-based intervention might then save money to the health care system, but put additional financial strain on the informal caregiver. We therefore strongly recommend that informal caregiver costs are included and a societal approach is adopted to assess the real cost-effectiveness of an intervention.

Include long-term costs and effects of treatments

The follow-up in the included studies was maximally twelve months after stroke onset. However, some late complications of stroke, such as depression only appear after one to two years. To ensure that all relevant costs and effects of interventions are included, we therefore recommend adopting a longer time horizon in cost-effectiveness studies for stroke interventions.

Adopt uniform measurement instruments
In order to compare studies, it is also advised to have more uniformity in the use of measurement instruments for health outcomes, e.g., Quality of Life and (social) participation scales.

Lastly, it was striking to see that even though some studies were published after 2005, all data analyzed were collected before 2000. It is questionable whether the results of these studies reflect current medical practice. We therefore recommend that new cost-effectiveness studies will be performed, investigating up-to-date ways of delivering integrated stroke care.

Reviewers

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Appendix. Methodological quality of the studies.

Author; year	Type of research (RCT, cohort)	Follow- up time	Patient series	Cost perspective	Clear description of intervention	Total	Comments
Anderson; 2000	++	+/-	+/-	++	+/-	**Good**	Only 6 months of follow-up, societal perspective
Andersson; 2002	+/-	+	+/-	+/-	+/-	**Moderate**	1 year of follow-up, non randomized trial
Beech; 1999	++	+	+	+	+/-	**Good**	1 year of follow-up, health care perspective
Bowen; 1994	+/-	- -	+	-	+	**Low**	Non-randomized trial with historical controls, healthcare perspective, insufficient description of which costs included, follow-up unclear
Claesson; 2000	++	+	+	++	+	**Good**	1 year of follow-up, societal perspective
Fjaertoft; 2005	+	+	+	+/-	++	**Good**	Only health care costs included
Gladman; 1994	+	+	+	+/-	+/-	**Good**	Only health care costs included; but informed after indirect costs
Holmqvist; 1996	-	+	- -	+/-	+/-	**Low**	Pilot study, only 15 subjects
Hui; 1995	+	+/-	+/-	+/-	+	**Good**	Health care perspective, limited number of participants
Markle-Reid; 2011	+	+	+/-	++	+	**Good**	1 year of follow-up, societal perspective, limited number of subjects
McNamee; 1998	+	+/-	+/-	+/-	+	**Good**	Only health services perspective, and only 6 months of follow-up. Limited number of participants
Moodie; 2006	+/-	+/-	+	+/-	+	**Moderate**	Cohort study, health care perspective, only 6 months
Odderson; 1993	+/-	-	+/-	-	++	**Low**	Evaluation with before and after data of the implementation of a clinical pathway, only average charges for acute stroke care per year available
Patel; 2004	++	+	+	++	+	**Good**	1 year of follow-up, societal perspective
Roderick; 2001	+	+/-	+/-	+/-	+/-	**Moderate**	6 months of follow-up, health care perspective
Teng; 2002	+	-	+/-	+/-	+/-	**Moderate**	3 months follow-up, health care perspective
Van Exel; 2005	+	+/-	+	+/-	+	**Good**	Health care perspective, 6 months of follow-up.
Van Koch; 2000	++	+	+/-	+/-	+/-	**Good**	Health care perspective, 1 year follow-up, RCT; detailed description elsewhere
Wentworth; 1996	+/-	-	+	-	+	**Low**	Only in-hospital costs included, 7 days of follow-up average, evaluation with before and after data of the implementation.

++, very good; +, good; +/-, moderate; - bad; -- very bad.