Annals of Physical and Rehabilitation Medicine 67 (2024) 101824

Available online at
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com

Original article

Stroke rehabilitation pathways during the first year: A cost-effectiveness analysis from a cohort of 460 individuals

Pedro Maciel Barbosa[a,b,c,*], Helena Szrek[d], Lara Noronha Ferreira[e,f,g], Vitor Tedim Cruz[b,h], João Firmino-Machado[h,i,j]

[a] Instituto de Saúde Pública, Universidade do Porto, Rua das Taipas 135, 4050-600 Porto, Portugal
[b] Hospital Pedro Hispano, Unidade Local de Saúde, EPE, Rua de Alfredo Cunha 365, 4450-021 Matosinhos, Portugal
[c] Centro de Investigação em Reabilitação, Escola Superior de Saúde, Instituto Politécnico do Porto, R. Dr. António Bernardino de Almeida, 4200-072 Porto, Portugal
[d] Centre for Economics and Finance, University of Porto, R. Dr. Roberto Frias, 4200-464 Porto, Portugal
[e] ESGHT, Universidade do Algarve, Estr. da Penha 139, 8005-246 Faro, Portugal
[f] Centre for Health Studies and Research of the University of Coimbra, Avenida Dias da Silva 165, 3004-512 Coimbra, Portugal
[g] Research Centre for Tourism, Sustainability and Well-Being (CinTurs), Portugal
[h] EPIUnit, Instituto de Saúde Pública, Universidade do Porto, Rua das Taipas 135, 4050-600 Porto, Portugal
[i] Centro Académico Clínico Egas Moniz, 810-193 Aveiro, Portugal
[j] Centro Hospitalar Vila Nova de Gaia/Espinho, R. Conceição Fernandes S/N, 4434-502 Vila Nova de Gaia, Portugal

ARTICLE INFO

Article History:
Received 3 July 2023
Accepted 17 January 2024

Keywords:
Pathway
Stroke
Rehabilitation
Outcomes
Cost-analysis
Health policy

ABSTRACT

Background: Stroke burden challenges global health, and social and economic policies. Although stroke recovery encompasses a wide range of care, including in-hospital, outpatient, and community-based rehabilitation, there are no published cost-effectiveness studies of integrated post-stroke pathways.
Objective: To determine the most cost-effective rehabilitation pathway during the first 12 months after a first-ever stroke.
Methods: A cohort of people in the acute phase of a first stroke was followed after hospital discharge; 51 % women, mean (SD) age 74.4 (12.9) years, mean National Institute of Health Stroke Scale score 11.7 (8.5) points, and mode modified Rankin Scale score 3 points. We developed a decision tree model of 9 sequences of rehabilitation care organised in 3 stages (3, 6 and 12 months) through a combination of public, semi-public and private entities, considering both the individual and healthcare service perspectives. Health outcomes were expressed as quality-adjusted life years (QALY) over a 1-year time horizon. Costs included healthcare, social care, and productivity losses. Sensitivity analyses were conducted on model input values.
Results: From the individual perspective, pathway 3 (Short-term Inpatient Unit » Community Clinic) was the most cost-effective, followed by pathway 1 (Rehabilitation Centre » Community Clinic). From the healthcare service perspective, pathway 3 was the most cost-effective followed by pathway 7 (Outpatient Hospital » Private Clinic). All other pathways were considered strongly dominated and excluded from the analysis. The total 1-year mean cost ranged between €12104 and €23024 from the individual's perspective and between €10992 and €31319 from the healthcare service perspective.
Conclusion: Assuming a willingness-to-pay threshold of one times the national gross domestic product (€20633/QALY), pathway 3 (Short-term Inpatient Unit » Community Clinic) was the most cost-effective strategy from both the individual and healthcare service perspectives. Rehabilitation pathway data contribute to the development of a future integrated care system adapted to different stroke profiles.

Introduction

Stroke rehabilitation should be intensive, timely, and multidisciplinary, with coordinated transfers between settings, and effective interfacing with social and community care [1]. However, post-stroke care is considered disorganized and fragmented even in the best health systems [2], and many survivors express dissatisfaction [3].

Abbreviations: C, Outpatient Community Clinic; H, Outpatient Day Hospital; HB, Home-based Rehabilitation; ICER, Incremental Cost-Effectiveness Ratio; MMSE, Mini Mental State Examnation; mRS, modified Rankin Scale; N, Nursing Home; NHS, National Health System; NIHSS, National Institute of Health Stroke Scale; P, Outpatient Private Clinic; QALY, Quality-Adjusted Life Year; RC, Rehabilitation Inpatient Centre; UL, Long-Term Inpatient Unit; UM, Medium-term Inpatient Unit; US, Short-term Inpatient Unit; WHO, World Health Organization; WTP, willingness-to-pay

* Corresponding author.
E-mail addresses: pmb@ess.ipp.pt (P.M. Barbosa), lnferrei@ualg.pt (L.N. Ferreira).

https://doi.org/10.1016/j.rehab.2024.101824

There are no universally agreed best practices across European countries, where different health service structures and payment systems often lead to inconsistent care pathways, type, and quality of therapies [4,5].

Stroke defies worldwide health, social and economic policies, as a global leading cause of mortality and disability [6]. By 2047 there will be an additional 40,000 incident stroke cases (+3 %) and 2.58 million prevalent stroke cases (+ 27 %) [7], and the corresponding economic burden will increase [8]. Approximately 3 to 4 % of total health expenditures in Western countries are allocated to stroke [9]. In 2017, 32 European countries spent €60 billion on stroke care, of which 45 % was for health care, 8 % for social care, 47 % for direct and indirect productivity losses. Of the €27 billion spent on stroke healthcare, only 18 % was spent on rehabilitation [10].

Despite recovery from a stroke being an arduous journey that takes months or years [11] and covers a whole spectrum of care, including in-hospital, outpatient, and community-based rehabilitation [12], there are no published cost-effectiveness studies of integrated post-stroke pathways [13]. Our previous work showed 9 different rehabilitation pathways, that suggested heterogeneity, inefficiency, and inequalities, followed by a heterogeneous satisfaction level [14].

In the last years, there has been a call to design new care settings that ensure greater consistency and effectiveness to meet the targets set for 2030 by the *Action Plan for Stroke* [4]. The extent of the challenge, combined with limited healthcare budgets emphasizes the need for an evidence-based and cost-effective analysis that contributes to post-stroke decision-making [15], bridging the gap between clinical guidelines, organizational models, and the provision of care.

This study evaluated the cost-effectiveness of post-stroke rehabilitation pathways using high-quality individual participant-level data. It aimed to (i) determine the cost structure of each pathway and ii) determine the most cost-effective rehabilitation post-stroke care pathway from the perspective of the individuals with stroke and that of the healthcare service.

Methods

Stroke rehabilitation pathways in Portugal

Portugal has a healthcare system based on a National Health System (NHS), with 8 rehabilitation care settings described as follows: Short-term Inpatient Unit (US), Medium-term Inpatient Unit (UM), Long-Term Inpatient Unit (UL), Rehabilitation Inpatient Centre (RC), Home-based Rehabilitation (HB), Outpatient Day Hospital (H), Outpatient Community Clinic (C), Outpatient Private Clinic (P), Nursing Home (N) [16]. A detailed description of outcomes and process level data for each set is provided in Appendix A1 using the following 6 variables: 1) admission assessment, 2) discharge plan, 3) stroke survivor involvement in the definition of rehabilitation goals, 4) multidisciplinary team available, 5) frequency and 6) duration of clinical sessions.

Considering the care circuits observed over the 4 follow-up stages (first 7 days, 3, 6 and 12 months after stroke), the 9 most common pathways were selected for this study, using the sequences of the 8 different rehabilitation settings mentioned above through a combination of public, semi-public and private organisations over the first year post-stroke. In each follow-up assessment, participants were asked about the setting where the rehabilitation had taken place since the last assessment, building up a multistep pathway of 4 rehabilitation settings. Graphic presentation and outcome descriptions by pathway are available in Appendix A2, and further details are reported elsewhere [16].

Model overview

We estimated the cost-effectiveness of the pathways from the individual and the healthcare service perspective during the first 12 months after stroke in Portugal. This was a secondary analysis that considered all the participants from a prospective cohort derived from our previous work [14,16]. It included a total of 460 individuals in the acute phase of a first stroke, 418 (90 %) of whom were followed up to 12 months (Fig. 1). In total, 51 % of participants were women, mean (SD) age was 74.4 (12.9) years, mean severity score was 11.7 (8.5) (NIHSS - National Institute of Health Stroke Scale), mode disability score was 3 (mRS – Modified Rankin Scale) and mean neurocognitive function score was 24.2 (3.1) (MMSE - Mini-Mental State Examination). After admission to a large public hospital in Portugal, individuals were screened for the following eligibility criteria: a) diagnosis of stroke confirmed by clinicians and supported by neuroimaging, b) first episode of stroke, and c) not globally aphasic. The study sample was restricted to individuals who could complete the participant-reported outcome measure - Health-related Quality of Life (EQ-5D-3 L) independently. The remaining outcomes (NHISS, mRS, MMSE), health benefits and costs were retrieved from person-level data of our cohort study and when unavailable national data sources were considered, as described in detail in Appendix A3. All participants signed informed consent and the study was approved by the ethics and data protection committee (No. 57/14/RS). Further details of the sample are reported elsewhere [14,16].

A decision tree model was used to analyse cost-effectiveness, considering individual participant-level data retrieved from our previous cohort study. Since post-stroke recovery-related processes are time-dependent, the Stroke Roundtable Consortium designate 6 months as the start of the chronic phase, when spontaneous recovery is usually at its limit [17]. Therefore, this study considered a 1-year time horizon, so that the cost-effectiveness ratio included the benefits of the main functional gains and utilities. This model considered the following outcomes: i) cost per quality-adjusted life year (QALY), ii) cost per pathway, and iii) cost per stroke survivor. We defined 9 competing pathways for our cost-effectiveness model (Fig. 1): 1: RC-C-C-C; 2: US-C-C-C; 3: RC-UM-C-C; 4: US-UM-C-C; 5: UM-UM-C-C; 6: UM-UM-HB-C; 7: H-H-C-C; 8: H-H-P-P; and 9: UL-UL-UL-NH. The tree model was implemented and analysed in TreeAgePro 2023. This manuscript conforms to the Consolidated Health Economic Evaluation Reporting Standards 2022 (CHEERS2022).

Health benefits

The health benefits considered in the decision model are summarized in Table 1 and their calculation is detailed in Appendices A4 and A5. The initial hospital sample was distributed as follows for each pathway: 8 % for pathway 1, 8 % for pathway 2, 10 % for pathway 3, 11 % for pathway 4, 18 % for pathway 5, 19 % for pathway 6, 7 % for pathway 7, 8 % for pathway 8 and 11 % for pathway 9.

The 3-month, 6-month, and 1-year relative survival estimates and utility values assigned to each health state of the decision model were obtained from previously published national studies [18,19]. A first-ever-in-a-lifetime stroke in Portugal was considered to have 1-year relative survival rate of 0.73; 6-month relative survival of 0.77; and, a 3-month relative survival of 0.81 [18,19]. These parameters varied by 10 % in the sensitivity analysis, to consider the variation in stroke epidemiology over time.

Utility values for the different health states ranged from a minimum of −0.324 to a maximum of 0.616, representing a health state value as worse than death and a value as good health, respectively [20]. All pathways presented positive 1-year mean utility values, except for pathway 9. Although in the Portuguese EQ-5D, tariff utility values for each health state range from −0.497 to 0.766 [20], negative values were considered as 0 in the decision model (Appendix A4).

Stroke survivors were considered to have a 1-year mean utility value of 0.388 for pathway 1, 0.438 for pathway 2, 0.242 for pathway 3, 0.285 for pathway 4, 0.041 for pathway 5, 0.021 for pathway 6, 0.166 for pathway 7, 0.177 for pathway 8, and −0.231 for pathway 9.

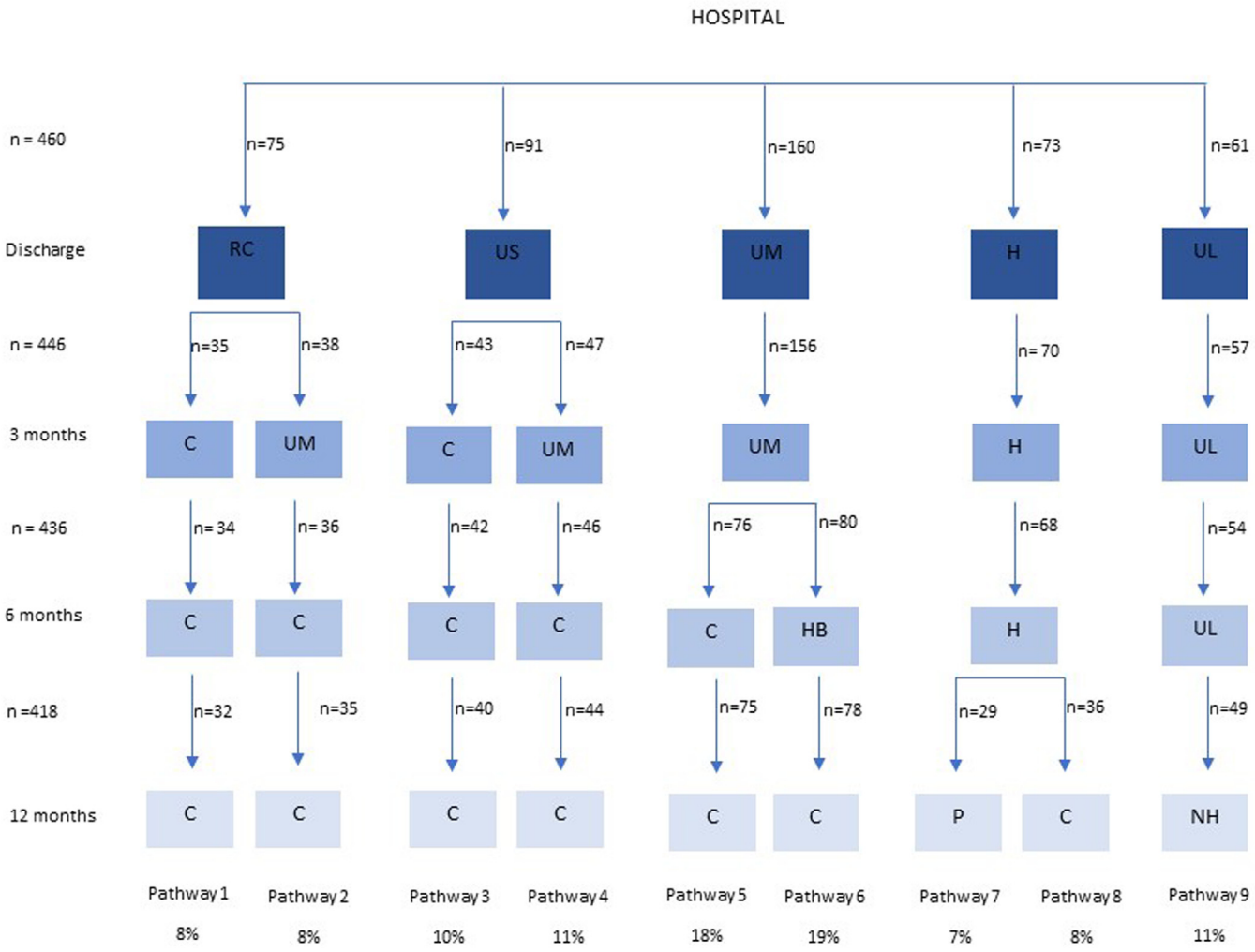

Fig. 1. Decision tree model overview. The figure depicts the 9 competing pathways of the cost-effectiveness model, during the first 12 months after a first-ever stroke. The transition probability between steps was defined as 1. The difference between the initial and final sample of each pathway is caused by follow-up losses by dropout or death: pathway 1 - 5 losses; pathway 2 - 3 losses; pathway 3 - 4 losses; pathway 4 - 3 losses; pathway 5 - 3 losses; pathway 6 - 4 losses; pathway 7 - 4 losses; pathway 8 - 4 losses; pathway 9 - 12 losses. Briefly, each pathway was as follows: 1 – Rehabilitation care at a Rehabilitation Centre from discharge to 3 months and at a Community Clinic from 3 to 12 months. 2 – Rehabilitation care at a Rehabilitation Centre from discharge to 3 months, at a Medium-term Unit from 3 to 6 months and at Community Clinic from 6 to 12 months. 3 – Rehabilitation care at a Short-term Unit from discharge to 3 months, and at Community Clinic from 3 to 12 months. 4 – Rehabilitation care at a Short-term Unit from discharge to 3 months, at a Medium-term Unit from 3 to 6 months and at Community Clinic from 6 to 12 months. 5 – Rehabilitation care at a Medium-term Unit from discharge to 3 months and at Community Clinic from 3 to 12 months. 6 – Rehabilitation care at a Medium-term Unit from discharge to 3 months, at Home-based from 3 to 6 months, and at Community Clinic from 6 to 12 months. 7 – Rehabilitation care at a Day Hospital from discharge to 6 months and at a Community Clinic from 6 to 12 months. 8 – Rehabilitation care at a Day Hospital from discharge to 6 months and at a Private Clinic from 6 to 12 months. 9 – Rehabilitation care at a Long-Term Unit from discharge to 6 months and at Nursing Home from 6 to 12 months. The percentages refer to the distribution of the initial hospital sample in each pathway.C - Community Clinic; H - Hospital; HB - Home-based; P - Private Rehabilitation; RC - Rehabilitation Centre; UL - Long-Term Unit; UM - Medium-term Unit; US - Short-term Unit.

The minimum and maximum benefit values observed for each individual in a certain pathway were used as range values in the sensitivity analysis (Table 1 and Appendix A5). The 1-year-benefits were retrieved from utility data determined in the same year. Therefore, the decision model did not incorporate discounts over time, as recommended by the World Health Organization (WHO) [21].

Costs

Costs were calculated assuming the following 2 independent payer perspectives: i) the healthcare service perspective, which includes only the costs and effects for the NHS part; ii) the individual perspective, which includes the cost and the effects for the individual with stroke. The underlying concepts of NHS and stroke costs followed Drummond et al. [22] and guidelines on good research practices [23].

Both individual and healthcare service perspective costs include 2 main domains – health and social costs (Appendix A6). Although the cost items for NHS and individual costs are the same, the point of view is different. The stroke rehabilitation pathways have important effects on costs across the healthcare service or the individual. There are items whose cost is fully supported by the healthcare service, others with costs shared between the healthcare service and the individual, and others that are paid totally by the individual. Each cost fraction was assigned to each perspective item by item. A detailed description of the cost calculation details by both perspectives is provided in Appendix A3.

Health costs comprise health intervention costs and individual transport costs. Social costs include social service costs (e.g. formal caregiver, day centre, nursing home), technical aid costs (e.g. walker, crutches, tripod, adjustable bed, recliner chair, wheelchair, shower chair, house adaptation for reduced mobility), and individual and informal caregiver productivity losses.

Table 1
Summary of decision tree model parameters.

Description of cost parameters By Stroke Survivor Perspective	Base case (€) [α]	Range (€) [a]	Further Details
Pathway 1	12,104	7672; 27,833	Includes Health (health care + transport) and Social (social care + technical aids + productivity losses) Costs. Please see Supplementary Appendices 3 and 6 for calculation details.
Pathway 2	16,537	10,352; 29,323	
Pathway 3	12,532	8437; 23,093	
Pathway 4	15,409	7679; 27,064	
Pathway 5	19,950	12,104; 35,960	
Pathway 6	19,902	10,844; 33,665	
Pathway 7	16,336	7792; 30,677	
Pathway 8	23,024	13,598; 36,897	
Pathway 9	21,856	12,877; 39,048	
Description of cost parameters By Healthcare Service Perspective	**Base case (€) [α]**	**Range (€) [a]**	**Further Details**
Pathway 1	26,428	24,131; 36,617	Includes Health (health care + transport) and Social (social care + technical aids + productivity losses) Costs. Please see Supplementary Appendices 3 and 6 for calculation details.
Pathway 2	31,319	28,814; 38,378	
Pathway 3	15,124	11,659; 21,750	
Pathway 4	18,424	16,301; 21,750	
Pathway 5	25,567	18,896; 27,091	
Pathway 6	24,761	18,740; 36,670	
Pathway 7	10,992	8102; 36,765	
Pathway 8	11,623	6637; 23,177	
Pathway 9	14,857	15,189; 40,044	
Description of health state	**Base case (QALY/year) [β]**	**Range (QALY/year) [b]**	**Further Details**
Pathway 1	0,388	0,306; 0,417	Includes one-year Quality-adjusted survival for each competing rehabilitation pathway. See Supplementary Appendices 4 and 5 for benefits details.
Pathway 2	0,272	0,199; 0,297	
Pathway 3	0,438	0,354; 0,475	
Pathway 4	0,305	0,220; 0,317	
Pathway 5	0,041	0,004:0,091	
Pathway 6	0,021	0; 0,049	
Pathway 7	0,177	0,110; 0,211	
Pathway 8	0,166	0,131; 0,221	
Pathway 9	0	0	

QALY, quality-adjusted life years; α, considers Portuguesa data sources (appendices 3 and 6); β, considers previously published national cohort study (appendices 4 and 5).
[a] : The parameter to be varied in the sensitivity analysis is the minimum and maximum cost values observed for each pathway. All the costs were determined in the reference year and obtained from Portuguese data sources, so there was no correction for inflation or purchasing power.
[b] : The parameter to be varied in the sensitivity analysis is the minimum and maximum benefit values observed for each patient individual in each pathway. Benefits were retrieved from utility data determined in the same year; therefore, the decision model did not incorporate discounts over time.

Health and Social costs were calculated using official legal publications from the Ministry of Health/Social Security, which define the value of the health and social service, as well as the criteria for attributing the cost to the healthcare service payer perspective or the individual payer perspective [24–32]. The only cost items whose value did not result from official publications and legal rules were the cost of technical aids and the cost of the Private setting, for which a market study was carried out based on the cost of 4 different providers.

Each pathway involved individuals with different health and social needs, so that, the minimum and maximum costs observed for each individual in a given pathway were used as range values in the sensitivity analysis (Table 1 and Appendix A6). The year considered for all cost calculations was 2021. All the costs were determined in the reference year and obtained from Portuguese data sources, so there was no correction for inflation or purchasing power, as proposed by the WHO [21].

Cost-effectiveness analysis

The incremental cost-effectiveness ratio (ICER) was calculated as the additional costs divided by the additional health benefits of one strategy compared to the next less-costly strategy. The ICERs were compared to the 2021 Portuguese gross domestic product per capita (ie, €20873), which is considered the external willingness-to-pay (WTP) threshold to identify the most cost-effective pathway. The "base case" was considered the non-dominated pathway with the lowest cost, ie, the reference strategy against which the other pathways in the model were compared. Reporting of the cost-effectiveness analysis followed the Consolidated Health Economic Evaluation Reporting Standards [23].

Results

The 1-year costs and benefits as cost-effectiveness results are presented in Table 2 and Appendices A5 and A6.

One-year costs and benefits

From the individual perspective, the total 1-year mean cost was lowest for pathway 1 and highest for pathway 9, ranging from €12104 to €21856, respectively. Health costs accounted for 24 % of the total costs in pathway 8, and <9 % in pathways 1, 3, 5, 6, 7 and 9. Pathways 1, 3 and 7 are free, whereas pathway 8 has costs >€4700 per survivor. Transportation costs range from €670 to 820 for pathways 1, 2, 3, 4, 7 and 8, and €0 for all the other pathways. Social costs ranged from 76 % pathway 8 to 95 % pathway 7 of the total cost. Stroke-related social care expenditure was about €1000 in pathways 5, 6 and 9, and varied between €500 to €800 in the other pathways. Loss of productivity costs ranged from €10,000 per individual in pathways 1 and 3, and up to €18000 in pathway 9.

From the healthcare service perspective, the total 1-year mean cost ranged from a minimum of €10,992 for pathway 7 to a maximum of €31319 for pathway 2. From this perspective, the relative weight of health costs exceeded 75 % of the total cost in pathways 1

Table 2
Costs, benefits, and incremental cost-effectiveness ratios, by pathway, from a stroke survivor and healthcare service perspective.

	1-year costs and benefits		
	Incremental Costs (€)	Incremental Benefits (QALY)	ICER (cost/QALY) [a]
Societal Perspective			
Pathway 1	–	–	–
Pathway 3	428	0,05	8560
Pathway 4	2877	−0,13	SD
Pathway 7	3804	−0,26	SD
Pathway 2	4005	−0,17	SD
Pathway 6	7370	−0,42	SD
Pathway 5	7418	−0,40	SD
Pathway 9	9324	−0,44	SD
Pathway 8	10492	−0,27	SD
Healthcare Service			
Pathway 7	–	–	–
Pathway 8	1531	−0,01	SD
Pathway 9	4765	−0,18	SD
Pathway 3	5032	0,26	19279,69
Pathway 4	3300	−0,13	SD
Pathway 6	9637	−0,41	SD
Pathway 5	10,443	−0,40	SD
Pathway 1	11,304	−0,05	SD
Pathway 2	16,195	−0,17	SD

ICERs were calculated using TreeAge software, which considers a larger number of decimal places than presented in the table. Therefore, the ICERs differ from calculations using the cost and QALY values are presented in the table.
ICER, incremental cost-effectiveness ratio; QALY, quality-adjusted life years; SD, strongly dominated.
[a] : The difference in cost divided by the difference in benefits of one pathway, compared with the next more effective pathway.

and 2, and 55 % in pathways 3, 4, 5, 6 and 9. Health care costs ranged from €1500 in pathways 7 and 8, to €20000 in pathways 1 and 2. Transport costs varied between €150 in pathway 9 and €1800 in pathway 5. Social costs ranged from 25 % of the total cost in pathways 1 and 2 to 85 % in pathway 8. Stroke-related social care expenditure was about €1300 in pathways 1 and 3, and €3800 in pathway 5. Loss of productivity costs ranged from €4000 in pathway 3, up to €7000 in pathway 9.

The quantification of QALYs showed that pathway 9 was the least effective, with −0,342 QALYs per individual over 1 year, whereas pathways 3 and 4 were the most effective, with 0,306 and 0,354 QALYs per individual, respectively.

Cost-effectiveness results

From the individual perspective, pathways 2 and 4 to 9 were strongly dominated by pathway 1, ie, they were costlier and less effective. Pathway 3 had an ICER of €8560 per QALY. From the healthcare service perspective, pathways 1, 2, 4, 5, 6, 8, and 9 were strongly dominated by pathway 7. Pathway 3 had an ICER of €19279 per QALY. All strongly dominated pathways were excluded from the analysis.

One-way sensitivity analysis

Sensitivity analysis results are detailed in Table 3. From the individual perspective, pathways 2, 4, 6, and 7 were not strongly dominated when the sensitivity analysis considered their minimal specific costs. Nevertheless, the most cost-effective pathways were always 1 and 3 for all the tested parameters. For the extreme cost value in pathway 1, along with the minimum cost value in pathway 3, the most cost-effective strategy was pathway 3. For all the other tested parameters, pathway 1 was the most cost-effective strategy.

From the healthcare service perspective, pathway 8 was not strongly dominated when the sensitivity analysis considered its minimal costs or maximum benefits. The most cost-effective pathways were always 7 and 3 for all the tested parameters. For minimum benefits and maximum cost of the pathway 3, the pathways 1 and 4 were considered undominated, respectively.

Discussion

Pathway 3 provides rehabilitation care to people with a first-ever stroke at a short-term inpatient unit (ie, from discharge to 3 months) followed by an outpatient community clinic from 3 to 12 months. This pathway was the most cost-effective strategy, both from the individual and healthcare service perspectives. Although at a national level, there is no universally accepted threshold for the WTP for a QALY, we interpreted the results according to the cut-offs commonly applied in cost-effectiveness analyses, which are set at one times the national gross domestic product per capita (ie, €20873 per QALY in Portugal in 2021).

However, policymakers may have short-term budget constraints that prevent them from adopting the most cost-effective pathway. Furthermore, the level of severity and clinical complexity of stroke is very varied, so a comprehensive rehabilitation provision model must include differentiated care pathways [1,7]. Pathway 1 from the stroke survivor perspective, and pathway 7 from the healthcare service's perspective, were both the least costly pathways and were still cost-effective in the final decision model, and therefore could be implemented.

From the individual perspective, pathways 3 and 1 were the most cost-effective, probably because both are free in terms of health care costs, have standardised transport costs, and the lowest social costs, as well as the largest health benefits. The sensitivity analysis showed that pathways 2, 4, 6 and 7 would be cost-effective if a minimum cost is assumed among the tested parameters. These results were

Table 3
Sensitivity analysis.

Parameter to be varied in the sensitivity analysis (stroke survivor perspective)		Parameter range [a]	Pathway [b]	ICER (societal perspective) [c]
Benefits Pathway 1 (QALY)	Minimum	0,306	1	–
			3	3242,42
	Maximum	0,417	1	–
			3	20380,95
Benefits Pathway 3 (QALY)	Minimum	0,354	1	–
			3	SD
	Maximum	0,475	1	–
			3	4919,54
Costs Pathway 1 (€)	Minimum	7672	1	–
			3	97200
	Maximum	27,833	3	–
			1	SD
Costs Pathway 3 (€)	Minimum	8437	3	–
			1	SD
	Maximum	23,093	1	–
			3	219780
Parameter to be varied in the sensitivity analysis (healthcare service perspective)		**Parameter range [a]**	**Pathway [b]**	**ICER (provider perspective) [c]**
Benefits Pathway 7 (QALY)	Minimum	0,11	7	–
			3	15341,46
			8	27339,29
	Maximum	0,211	7	–
			3	22167,40
Benefits Pathway 3 (QALY)	Minimum	0,354	7	–
			3	28429,38
			1	332470,59
	Maximum	0,475	7	–
			3	16885,91
Costs Pathway 7 (€)	Minimum	8102	7	–
			3	26904,21
	Maximum	21,292	7	–
			3	12871,32
Costs Pathway 3 (€)	Minimum	11,659	7	–
			3	6003,83
	Maximum	21,750	7	–
			3	44666,67
			4	65093,75

ICER, incremental cost-effectiveness ratio; QALY, quality-adjusted life years; SD, strongly dominated.
[a] : The table depicts the minimum and maximum values of the parameters assumed in the sensitivity analysis and the corresponding ICERs.
[b] : only undominated pathways were considered.
[c] : The difference in cost divided by the difference in benefits of one pathway, compared with the next more effective pathway. All the ICER values were computed using a cost-effectiveness model that assumes as an outcome the number of quality-adjusted life years.

obtained for very extreme values, which we consider unlikely to occur. However, they refer to a relevant aspect from the point of view of health policy and care pathway definition: the importance of costs in the final decision [10]. Indeed, in the case of an impactful health condition such as stroke, it is important to reflect on whether the choice and access to the optimal rehabilitation pathway should rely on cost considerations. Alternatively, decision-makers may fix a different threshold according to the severity of the stroke and the corresponding rehabilitation potential. These results are in line with other studies that highlight a lack of guidance to aid rehabilitation decision-making that is influenced by participant-level and organizational factors that occur across services and countries [33].

From the healthcare system perspective, the cost-effective pathways 3 and 7 are distinguished by their low healthcare costs compared to the pathways considered dominant. This difference is partly explained by the fact that pathways 3 and 7 involve a reduced exposure to inpatient settings, which are more expensive for the health service (NHS). However, evidence has shown the advantages of these settings for the rehabilitation of certain pathologies, including acute ischemic stroke [34,35]. Considering the health system's financial sustainability [36], it should be noted that all the pathways are currently operationally available but only 30 % were considered as cost-effective in our study. So, future healthcare planning should seek to define a more efficient rehabilitation system with a smaller number of pathways and more clearly defined, individualised pathways according to different stroke severity profiles. Analyses of health system efficiency determinants in the literature report that specific institutional arrangements should be avoided, namely gatekeeping and the fragmented presence of multiple healthcare services [37]. The existence of multiple care options may reduce health benefits and increase costs [38].

Our data show that stroke rehabilitation pathway cost-effectiveness analysis is consistent with evidence-based practice [39], with better incremental cost-effectiveness ratios assigned to pathways in which care settings offer more intensive and multidisciplinary rehabilitation [40]. We found that only the cost-effective pathways 1, 3 and 7, which started in Rehabilitation Inpatient Centres, Short-term Inpatient Units and Outpatient Day Hospital settings respectively, guarantee the recommended interdisciplinary and intense care. However, this care is restricted to 30 to 60 days, which means that only 30 % of the study sample was able to obtain such care. Beyond that, only 35 % of pathways include the individual in goal setting and only 60 % of pathways plan coordinated transitions. This underscores the necessity of funding more comprehensive and integrated care approaches.

This discrepancy between the care given and recommended good practice has been found in various European health systems and nations [41]. Two hypotheses may be suggested to explain this. The first is that professionals and health systems are not aware of the guidelines or have not integrated them systemically into their decision-making process [42]. The second is that the guidelines do not account for the process's complexity. These results demonstrate the need for policy approaches to overcome barriers to evidence-based stroke rehabilitation [43].

The current study provides the first detailed 1-year cost-effectiveness analysis of different multistep pathways of stroke rehabilitation. Models of integrated rehabilitation care and individual itineraries have been little reported in the literature. An econometric study evaluated an integrated care pathway for people after stroke, however, no alternatives were compared and the focus was only on outpatient community rehabilitation, with no evaluation after 6 months [44]. In contrast, there is plenty of literature analysing the cost-effectiveness of individual models of care, such as early supported discharge services [45], home rehabilitation [46], stroke units [34], and stroke services [35]; however, individuals' needs, expectations, and concerns are not translated into research. The main wish of people after stroke is to have timely access to health services and, when necessary, to be easily guided from one service to another, without obstacles, delays, or loss of information [47]. Therefore, research should develop study models that provide the data needed for decision-making and development of care provision.

The major strength of this study is that it essentially considered rehabilitation pathways using participant-level data drawn from a robust original cohort study [14,16]. The present study also adds to most previous cost-effectiveness analyses on this topic by including people with moderate and severe stroke, estimating costs and benefits from both the individual and health service perspectives, and involving a 1-year analysis [13]. It should be noted that the cost parameters were all obtained in Portugal in the reference year, which excludes the need for correction for purchasing power and inflation and increases the data robustness.

Study limitations

Although we recognise that the single-centre methodological design restricts external validity, we do not consider this to be a limitation, as the results are applicable in countries with similar organisational models. Moreover, it is acknowledged by the Action Plan for Stroke that new settings of care should be designed to meet the targets set for 2030 [5]. So, the present analysis of different integrated pathways of stroke rehabilitation is required for the widespread uptake of integrated care models and effective navigation through complex health systems [3,4]. The Portuguese stroke rehabilitation system, despite being fragmented, includes different models of care (inpatient, outpatient, and home-based) whose organisational characteristics are common to different countries and health systems. Data relating to the access of different severity and socio-demographic stroke profiles to different levels of care and their distribution within each level of care invite comparison and contribute to the international discussion about policies that enhance transitions across different environments [2]. However, some limitations of our study need to be addressed. Impairment of cognitive function may have reduced the accuracy of the information reported by the individuals with stroke, however, this phenomenon is not differential between the majority of sub-samples/pathways. Besides, only the EQ-5D-3L outcome was self-reported, so cognitive status is expected to have a minor contribution to information bias, with little impact on the internal validity of the results. Considering the stroke pathways, parameters were obtained from the cohort study that described 11 pathways; but, because of sample size limitations, the analysis was performed on the 9 most prevalent. This can be considered a selection bias that limits the full analysis of the healthcare system. Additionally, despite including a wide range of ages, stroke subtypes and severity grades, a stratified econometric analysis was not carried out, as we did not expect a substantial difference based on previous results of this cohort. On the other hand, as the Portuguese NHS is public, the results cannot be extrapolated to itineraries initially started from a private hospital. Furthermore, when compared with the 5L version, the EQ-5D-3L overestimates health problems, which consequently underestimates utilities and discriminatory power [48]. Future studies should consider the more complete version of the EQ-5D to increase the sensitivity and accuracy of benefit analysis. In addition, different pathways have shown different weightings of health and social costs, however, exploring the model from both the stroke survivor and healthcare service perspectives showed that this difference does not affect the conclusion about the most cost-effective intervention. Finally, the present study considers a timeframe of 1 year instead of a longer period. It would be important to consider longer follow-ups in future studies that evaluate the impact of long-term costs and benefits, such as the return to professional activity.

Conclusion

Our study provides evidence for policymakers to designate pathway 3 as the new standard-of-care pathway for stroke rehabilitation, under a WTP threshold of one times the national gross domestic product (€20873 per QALY). Given the complexity of the clinical heterogeneity of stroke, other pathways may be considered, such as pathway 1 from the individual perspective, and pathway 7 from the healthcare service perspective, which were still cost-effective in the final decision model. This allows the planning and organization of a comprehensive, integrated, and efficient system adapted to the different stroke profiles.

List of appendices

Appendix A1 - Settings of Care Description
Appendix A2 – Pathways
Appendix A3 - Cost Calculation Details
Appendix A4 – Utilities
Appendix A5 - Benefits_QALY
Appendix A6 - Cost Structure
Appendix A7 – Guidelines Checklist

Declaration of competing interest

None.

Acknowledgements

We are grateful to all the individuals with stroke who took part in the study and thank all primary care practices and physicians who collaborated with us.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Supplementary materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.rehab.2024.101824.

References

[1] Langhorne P, Sandercock P, Prasad K. Evidence-based practice for stroke. Lancet Neurol 2009;8(4):308–9. doi: 10.1016/S1474-4422(09)70060-2.
[2] Cameron JI, Tsoi C, Marsella A. Optimizing stroke systems of care by enhancing transitions across care environments. Stroke 2008;39(9):2637–43. doi: 10.1161/STROKEAHA.107.501064.
[3] McKee M, Merkur S, Edwards N, Nolte E. The changing role of the hospital in European health systems. Cambridge: Cambridge University Press; 2020.
[4] Stevens E, McKevitt C, Emmett E, Wolfe C, Wang Y. The burden of stroke in Europe report. London: Stroke Alliance for Europe; 2017.
[5] Norrving B, Barrick J, Davalos A, Dichgans M, Cordonnier C, Guekht A, et al. Action plan for stroke in Europe 2018-2030. Eur Stroke J 2018;3:309–36. doi: 10.1177/2396987318808719.
[6] GBD 2019 Diseases and Injuries Collaborators. Global burden of 369 diseases and injuries in 204 countries and territories, 1990-2019: a systematic analysis for the Global Burden of Disease Study 2019. Lancet 2020;396(10258):1204–22. doi: 10.1016/S0140-6736(20)30925-9.
[7] Wafa HA, Wolfe CDA, Emmett E, Roth GA, Johnson CO, Wang Y. Burden of stroke in Europe: thirty-year projections of incidence, prevalence, deaths, and disability-adjusted life years. Stroke 2020;51(8):2418–27. doi: 10.1161/STROKEAHA.120.029606.
[8] GBD 2016 Neurology Collaborators. Global, regional, and national burden of neurological disorders, 1990-2016: a systematic analysis for the Global Burden of Disease Study 2016. Lancet Neurol 2019;18(5):459–80. doi: 10.1016/S1474-4422(18)30499-X.
[9] Struijs JN, van Genugten ML, Evers SM, Ament AJ, Baan CA, van den Bos GA. Future costs of stroke in the Netherlands: the impact of stroke services. Int J Technol Assess Health Care 2006;22(4):518–24. doi: 10.1017/S0266462306051464.
[10] Luengo-Fernandez R, Violato M, Candio P, Leal J. Economic burden of stroke across Europe: a population-based cost analysis. Eur Stroke J 2020;5(1):17–25. doi: 10.1177/2396987319883160.
[11] Yeoh YS, Koh GC, Tan CS, Tu TM, Singh R, Chang HM, et al. Health-related quality of life loss associated with first-time stroke. PLoS One 2019;14(1):e0211493. doi: 10.1371/journal.pone.0211493.
[12] Allen D, Rixson L. How has the impact of 'care pathway technologies' on service integration in stroke care been measured and what is the strength of the evidence to support their effectiveness in this respect? Int J Evid Based Healthc 2008;6(1):78–110. doi: 10.1111/j.1744-1609.2007.00098.x.
[13] Tummers JF, Schrijvers AJ, Visser-Meily JM. Economic evidence on integrated care for stroke patients; a systematic review. Int J Integr Care 2012;12:e193. doi: 10.5334/ijic.847.
[14] Maciel Barbosa P, Firmino-Machado J, Ferreira LN, Tedim Cruz V, Szrek H. From healthcare system to individuals through stroke rehabilitation pathways. Outcomes, information, and satisfaction along 12 months prospective cohort in Portugal. Top Stroke Rehabil 2023;30(7):727–37. doi: 10.1080/10749357.2023.2165266.
[15] Earnshaw SR, Wilson M, Mauskopf J, Joshi AV. Model-based cost-effectiveness analyses for the treatment of acute stroke events: a review and summary of challenges. Value Health 2009;12(4):507–20. doi: 10.1111/j.1524-4733.2008.00467.x.
[16] Barbosa PM, Ferreira LN, Cruz VT, Silva A, Szrek H. Healthcare, clinical factors and rehabilitation predicting quality of life in first-time stroke patients: a 12-month longitudinal study. J Stroke Cerebrovasc Dis 2022;31(4):106300. doi: 10.1016/j.jstrokecerebrovasdis.2021.106300.
[17] Grefkes C, Fink GR. Recovery from stroke: current concepts and future perspectives. Neurol Res Pract 2020;2:17. doi: 10.1186/s42466-020-00060-6.
[18] Correia M, Magalhães R, Silva MR, Matos I, Silva MC. Stroke types in rural and urban northern Portugal: incidence and 7-year survival in a community-based study. Cerebrovasc Dis Extra 2013;3(1):137–49. doi: 10.1159/000354851.
[19] Abreu P, Magalhães R, Baptista D, Azevedo E, Silva MC, Correia M. Readmissions and mortality during the first year after stroke-data from a population-based incidence study. Front Neurol 2020;11:636. doi: 10.3389/fneur.2020.00636.
[20] Ferreira LN, Ferreira PL, Pereira LN, Oppe M. The valuation of the EQ-5D in Portugal. Qual Life Res 2014;23(2):413–23. doi: 10.1007/s11136-013-0448-z.
[21] WHO guide to cost-effectiveness analysis, http://www.who.int/choice/publications/p_2003_generalised_cea.pdf; 2003 [accessed 13 July 2023].
[22] Drummond MFS, Mark J, Claxton K, Stoddart GL, Torrance GW. Methods for the economic evaluation of health care programmes. 4th ed. Oxford: Oxford University Press; 2015.
[23] Husereau D, Drummond M, Augustovski F, de Bekker-Grob E, Briggs AH, Carswell C, et al. Consolidated Health Economic Evaluation Reporting Standards 2022 (CHEERS 2022) explanation and elaboration: a report of the ISPOR CHEERS II good practices task force. Value Health 2022;25. doi: 10.1016/j.jval.2021.10.008.
[24] Decreto-Lei n.101/2006. (Health cost definition by national law).
[25] Decreto-Lei n.37/2022. (Health cost definition by national law).
[26] Circular Normativa ACSS_03/2021. (Health cost definition by national law).
[27] Decreto-Lei n.93/2009. (Technical aids cost calculation).
[28] Decreto-Lei n.74/2022. (Technical aids cost calculation).
[29] Despacho n.7980-A/2022. (Transport cost definition by national law).
[30] Decreto-Lei n.74/2022. (Social cost definition by national law).
[31] Despacho n.7980-A/2022. (Social cost definition by national law).
[32] Decreto-Lei n.28/2004. (Productivity losses cost calculation).
[33] Longley V, Peters S, Swarbrick C, Bowen A. What factors affect clinical decision-making about access to stroke rehabilitation? A systematic review. Clin Rehabil 2019;33(2):304–16. doi: 10.1177/0269215518808000.
[34] Schmidt A, Heroum C, Caumette D, Le Lay K, Bénard S. Acute Ischemic Stroke (AIS) patient management in French stroke units and impact estimation of thrombolysis on care pathways and associated costs. Cerebrovasc Dis 2015;39(2):94–101. doi: 10.1159/000369525.
[35] Guzauskas GF, Boudreau DM, Villa KF, Levine SR, Veenstra DL. The cost-effectiveness of primary stroke centers for acute stroke care. Stroke 2012;43(6):1617–23. doi: 10.1161/STROKEAHA.111.648238.
[36] Liaropoulos L, Goranitis I. Health care financing and the sustainability of health systems. Int J Equity Health 2015;14:80. doi: 10.1186/s12939-015-0208-5.
[37] Hadad S, Hadad Y, Simon-Tuval T. Determinants of healthcare system's efficiency in OECD countries. Eur J Health Econ 2013;14(2):253–65. doi: 10.1007/s10198-011-0366-3.
[38] Berwick DM, Nolan TW, Whittington J. The triple aim: care, health, and cost. Health Aff 2008;27(3):759–69. doi: 10.1377/hlthaff.27.3.759.
[39] Langhorne P, Cadilhac D, Feigin V, Grieve R, Liu M. How should stroke services be organised? Lancet Neurol 2002;1(1):62–8. doi: 10.1016/s1474-4422(02)00011-x.
[40] Morii Y, Abiko K, Osanai T, Takami J, Tanikawa T, Fujiwara K, et al. Cost-effectiveness of seven-days-per-week rehabilitation schedule for acute stroke patients. Cost Eff Resour Alloc 2023;21(1):12. doi: 10.1186/s12962-023-00421-3.
[41] Teasell R, Mehta S, Pereira S, McIntyre A, Janzen S, Allen L, et al. Time to rethink long-term rehabilitation management of stroke patients. Top Stroke Rehabil 2012;19(6):457–62. doi: 10.1310/tsr1906-457.
[42] Juckett LA, Wengerd LR, Faieta J, Griffin CE. Evidence-based practice implementation in stroke rehabilitation: a scoping review of barriers and facilitators. Am J Occup Ther 2020;74(1) 7401205050p1-7401205050p14. doi: 10.5014/ajot.2020.035485.
[43] English C, Bayley M, Hill K, Langhorne P, Molag M, Ranta A, et al. Bringing stroke clinical guidelines to life. Int J Stroke 2019;14(4):337–9. doi: 10.1177/1747493019833015.
[44] Abdul Aziz AF, Mohd Nordin NA, Muhd Nur A, Sulong S, Aljunid SM. The integrated care pathway for managing post stroke patients (iCaPPS©) in public primary care Healthcentres in Malaysia: impact on quality adjusted life years (QALYs) and cost effectiveness analysis. BMC Geriatr 2020;20(1):70. doi: 10.1186/s12877-020-1453-z.
[45] Saka O, Serra V, Samyshkin Y, McGuire A, Wolfe CC. Cost-effectiveness of stroke unit care followed by early supported discharge. Stroke 2009;40(1):24–9. doi: 10.1161/STROKEAHA.108.518043.
[46] Candio P, Violato M, Luengo-Fernandez R, Leal J. Cost-effectiveness of home-based stroke rehabilitation across Europe: a modelling study. Health Policy 2022;126