CLINICAL AND POPULATION SCIENCES

High-Intensity Aphasia Therapy Is Cost-Effective in People With Poststroke Aphasia: Evidence From the COMPARE Trial

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BACKGROUND: Evidence from systematic reviews confirms that speech and language interventions for people with aphasia during the chronic phase after stroke (>6 months) improve word retrieval, functional communication, and communication-related quality of life. However, there is limited evidence of their cost-effectiveness. We aimed to estimate the cost per quality-adjusted life year gained from 2 speech and language therapies compared with usual care in people with aphasia during the chronic phase (median, 2.9 years) after stroke.

METHODS: A 3-arm, randomized controlled trial compared constraint-induced aphasia therapy plus (CIAT-Plus) and multimodality aphasia therapy (M-MAT) with usual care in 216 people with chronic aphasia. Participants were administered a standardized questionnaire before intervention and at 12 weeks after the 2-week intervention/control period to ascertain health service utilization, employment changes, and informal caregiver burden. Unit prices from Australian sources were used to estimate costs in 2020. Quality-adjusted life years were estimated using responses to the EuroQol-5 Dimension-3 Level questionnaire. To test uncertainty around the differences in costs and outcomes between groups, bootstrapping was used with the cohorts resampled 1000 times.

RESULTS: Overall 201/216 participants were included (mean age, 63 years, 29% moderate or severe aphasia, 61 usual care, 70 CIAT-Plus, 70 M-MAT). There were no statistically significant differences in mean total costs ($13 797 usual care, $17 478 CIAT-Plus, $11 113 M-MAT) and quality-adjusted life years (0.19 usual care, 0.20 CIAT-Plus, 0.20 M-MAT) between groups. In bootstrapped analysis of CIAT-Plus, 21.5% of iterations were likely to result in better outcomes and be cost saving (dominant) compared with usual care. In contrast, 72.4% of iterations were more favorable for M-MAT than usual care.

CONCLUSIONS: We observed that both treatments, but especially M-MAT, may result in better outcomes at an acceptable additional cost, or potentially with cost savings. These findings are relevant in advocating for the use of these therapies for chronic aphasia after stroke.

GRAPHIC ABSTRACT: A graphic abstract is available for this article.

Key Words: aphasia ■ health care cost ■ language therapy ■ rehabilitation ■ speech ■ stroke

Aphasia, a language disability affecting the ability to communicate, affects ≈34% of individuals with stroke in the chronic phase.[1] In a 2004 study using Medicare data in the United States of America, it was observed that aphasia was associated with an additional cost of US$1703 over 1-year poststroke relative to no aphasia, with differences primarily related to nursing home costs.[2] There are several interventions for people with aphasia that improve functional communication,[3] but further research is required to determine which

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Nonstandard Abbreviations and Acronyms

CIAT-Plus	constraint-induced aphasia therapy plus
EQ-5D-3L	EuroQol-5 Dimension-3 Level
M-MAT	multimodality aphasia therapy
QALY	quality-adjusted life year

treatments lead to better patient outcomes and reduced health care costs.

In particular, there are limited data on service use by stroke survivors with aphasia and on the cost-effectiveness of therapies provided for people with aphasia, particularly in the chronic phase after stroke. Economic evaluations are used to compare the costs and outcomes between one or more options to support informed decision-making. These studies have a role in understanding whether the therapies are cost-effective (ie, improvements in outcomes from therapies are gained at an acceptable additional cost). Few economic evaluations of speech and language therapies for people with stroke have been published.[4–7] In these studies, there was limited evidence that speech and language therapy was cost-effective compared with standard care.

In the randomized controlled trial of constraint-induced or multimodality personalised aphasia rehabilitation (COMPARE), undertaken in Australia and New Zealand, 2 therapies that have previously been shown to improve verbal communication, constraint-induced aphasia therapy plus (CIAT-Plus)[8] and multimodality aphasia therapy (M-MAT),[9] were compared with usual care for people with chronic (>6 months) poststroke aphasia.[10] To improve verbal communication, CIAT-Plus uses verbal and orthographic therapist-delivered cueing while M-MAT additionally includes multimodal tasks and cues (drawing, gesturing, and writing). Both are intensive, high-dose interventions delivered in a small group setting of 2 to 3 participants. Usual care did not involve any trial therapy. When compared with usual care, it was found that CIAT-Plus and M-MAT improved word retrieval based on the COMPARE naming battery, and self-reported functional communication and aphasia-related quality of life measures.[10] A cost-effectiveness analysis was planned as part of the trial as a secondary objective. In this study, we aimed to estimate the cost-effectiveness of CIAT-Plus and M-MAT when compared with usual care.

METHODS

COMPARE was a 3-arm, multicenter, parallel-group, open-label, blinded end point, phase III randomized controlled trial.[10,11] A data safety and management committee comprising staff independent of the principal investigators (a biostatistician, an experienced stroke rehabilitation trialist, and a speech pathologist with additional expertise in bioethics) monitored study progress and safety. The data that support the findings of this study are available from the corresponding author upon reasonable request.

Two intervention arms (CIAT-Plus[8] and M-MAT[9]) were compared with usual care. In the COMPARE study, the small group sessions for the 2 intervention groups were provided by 30 qualified and study-trained speech pathologists in community settings in 10 cities across Australia and New Zealand. The intervention was provided 3 hours per day, 5 days per week for 2 weeks (30 hours of treatment in total). In addition, a daily home practice communication task (15 minutes) was prescribed for each participant and checked for completion and logged the following day. usual care did not involve any trial therapy and comprised any therapy that was already accessed by participants. Approximately one-third of participants in all arms of the study had aphasia care other than the trial therapy.[10]

Participants in COMPARE were adults (18 years of age or older) living in the community who had been diagnosed with any type of aphasia (Western Aphasia Battery—Revised aphasia quotient[12] <93.8) following a stroke at least 6 months before enrollment in the trial. Participants were recruited through 22 hospitals with ethical approval for the study and direct community advertising in Australia and New Zealand. Consented participants were randomized to one of the 3 study groups in a 1:1:1 ratio, stratified by their aphasia severity (Western Aphasia Battery—Revised aphasia quotient mild=93.7–62.6; moderate=62.5–31.3; severe<31.3) with a central allocation system using blocked randomization within each stratum. Of the 216 people recruited, 21 were from New Zealand.

The trial was registered at the Australian New Zealand Clinical Trials Registry (https://www.clinicaltrials.gov; Unique identifier: 12615000618550) on June 15, 2016. The final follow-up data collection was completed on 23/07/2020.

Economic Evaluation

The Consolidated Health Economic Evaluation Reporting Standard, which are guidelines for reporting economic evaluations, was followed for this study.[13] The economic evaluation was prospectively planned from the outset of the trial.[10,11] A separate economic evaluation protocol was not published.

Health service use and productivity impacts (changes to employment and informal caregiving) were captured using a standardized questionnaire administered at baseline and 12 weeks following completion of the 2-week intervention or usual care (control) period (Supplemental Material). At baseline, information was obtained about employment status and health service use before study participation. At the 12-week follow-up, participants were asked about health service use in the time since the baseline assessment, informal caregiver burden, and changes in employment (participant and carer). For participants who reported the use of health services, additional information was obtained about the occasions and types of health services used within several predefined categories (eg, hospital presentations, inpatient rehabilitation, general practitioner visits). Where participants' communication disabilities prevented reporting this information, their family/friend answered questions on their behalf.

Costs were estimated from a societal perspective with the main focus on the health care sector and expressed in

Australian dollars (AUD). No discounting was undertaken since the duration of the follow-up was <12 months. Costs from a community health care perspective were also presented. Unit prices from Australian sources were obtained and inflated to the equivalent price in 2020 using the Total Health Price Index published by the Australian Institute of Health and Welfare[14] (Table 1). Informal care (defined as help received from family or friends to help them stay in their home including transport, cooking, cleaning, shopping, and bathing), and changes to employment for participants and carers were included as indirect costs when estimating broader societal costs in addition to the health sector costs. Costs involved in the delivery of the intervention were AUD 2000 per therapist for 30 hours of intervention per group (at 3 participants per group) equating to AUD 22.22 per hour per participant (AUD ≈600 total). Conversion to the US dollar equivalent can be made by multiplying prices/costs by 1.454 (purchasing power parity).

The primary outcome of the economic evaluation was the cost per quality-adjusted life year (QALY) gained in CIAT-Plus or M-MAT compared with usual care. A QALY is a metric that comprises both the length and quality of life, with one QALY being the equivalent of 1 year of healthy life. QALYs were estimated using responses to the EuroQol-5 Dimension-3 Level (EQ-5D-3L) questionnaire administered at baseline and 12-week follow-up. The EQ-5D-3L is a patient-reported outcome measure with questions related to mobility, self-care usual activities, pain/discomfort, and anxiety/depression. Responses were converted to a utility value using an algorithm validated in an Australian general population.[21] The EQ-5D-3L questionnaire is thought to be less sensitive to communication-related changes to quality of life than the Stroke and Aphasia Quality of Life Scale-39.[10] However, since responses to the Stroke and Aphasia Quality of Life Scale-39 have not been valued to estimate utility values, it could not be used to estimate QALYs for this analysis.

The cost per QALY gained was also estimated with costs limited to community health care thought to most likely be affected by the interventions (general practitioner, speech pathology, social work, psychologist, psychiatrist, neurologist, home rehabilitation, community-based aphasia, aids and equipment, and private speech therapy). A cost-effectiveness threshold of AUD 50 000 per QALY gained was used.

Statistics

Between-group differences were assessed using χ^2 tests for categorical variables, and Student *t* tests (mean) and Kruskal-Wallis tests (median) for continuous variables. A $P<0.05$ was considered to be statistically significant. Multiple imputation

Table 1. Unit Prices

	Unit price (AUD)	Units	Data source
Trial intervention	22.22	Per hour	Trial investigators
Hospital presentations			
ED admission	732.00	Per visit	Independent Hospital Pricing Authority[15]
In-patient admissions	2285.00	Per day	Independent Hospital Pricing Authority[15]
Inpatient rehabilitation	1010.77	Per day	Independent Hospital Pricing Authority[15]
Outpatient rehabilitation	332.00	Per service	Independent Hospital Pricing Authority[15]
General practitioner	52.00	Per visit	Australian Government, Department of Health[16]
Allied health services	63.25	Per session	Australian Government, Department of Health[16]
Psychiatrist	414.22	Per visit	Freed et al[17]
Neurologist	414.22	Per visit	Freed et al[17]
Other health services*	13.70–414.22		Several sources including the Australian Government[16] and Freed et al[17]
Home and community services			
Aphasia support	68.93	Per session	Trial investigators
Nursing services	64.03	Per visit	Farag et al[18]
Anticoagulation therapy	13.70	Per test	Australian Government, Department of Health[16]
Delivered meals	12.67	Per meal	Farag et al[18]
Personal care	41.55	Per hour	Farag et al[18]
Housework help	44.60	Per hour	Farag et al[18]
Gardening/home maintenance	59.66	Per service	Farag et al[18]
Home respite	1168.00	Per day	Private-in-home care cost, 2020 (www.daughterlycare.com.au)
Patient transport	14.14	Per trip	Farag et al[18]
Aids, equipment, home modifications	…		Patient report in this trial
Private speech therapy	145.00	Per session	Study investigators
Respite care	56.70	Per day	Australian Government, Department of Health and Aged Care[19]
Employment (patient and carer)	…		Patient report in this trial
Informal care	36.12	Per hour	Deloitte Access Economics[20]

Prices were adjusted to the 2020 Australian dollar equivalent (AUD).
*See Table S2.

Table 2. Resource Utilization to 12 Weeks

		Usual care N=61	CIAT N=70	M-MAT N=69
Trial intervention therapy, h	Mean (SD)	…	28 (4)	28 (5)
Emergency department attendance	n (%)	10 (16)	6 (9)	11 (16)
No. of visits	Mean (SD)	1 (1)	1 (0)	1 (0)
Hospital admission	n (%)	10 (16)	9 (13)	2 (3)
No. of visits	Mean (SD)	1 (0)	1 (0)	1 (0)
Length of stay	Mean (SD)	4 (6)	8 (8)	9 (6)
Rehabilitation admissions	n (%)	1 (2)	3 (4)	0 (0)
No. of visits	Mean (SD)	1 (-)	2 (1)	…
Length of stay	Mean (SD)	13 (-)	7 (7)	…
Outpatient rehabilitation	n (%)	15 (25)	22 (31)	22 (32)
No. of services	Mean (SD)	33 (74)	21 (19)	24 (41)
General practitioner	n (%)	48 (79)	51 (73)	55 (80)
No. of visits	Mean (SD)	3 (2)	2 (2)	2 (2)
Speech pathology	n (%)	3 (5)	1 (1)	2 (3)
No. of visits	Mean (SD)	3 (1)	5 (-)	8 (6)
Private speech therapy	n (%)	9 (15)	11 (16)	10 (14)
No. of sessions	Mean (SD)	16 (8)	8 (6)	10 (9)
Social work	n (%)	2 (3)	0 (0)	0 (0)
No. of visits	Mean (SD)	3 (2)	…	…
Psychologist	n (%)	3 (5)	3 (4)	2 (3)
No. of visits	Mean (SD)	2 (1)	7 (6)	2 (0)
Psychiatrist	n (%)	2 (3)	0 (0)	0 (0)
No. of visits	Mean (SD)	1 (0)	…	…
Neurologist	n (%)	8 (13)	12 (17)	3 (4)
No. of visits	Mean (SD)	2 (1)	1 (0)	1 (0)
Other health services*	n (%)	32 (52)	37 (53)	25 (36)
No. of visits	Mean (SD)	5 (8)	5 (6)	5 (6)
Home rehabilitation	n (%)	6 (10)	15 (21)	11 (16)
No. of visits	Mean (SD)	10 (7)	30 (52)	25 (30)
Community based aphasia	n (%)	20 (33)	20 (29)	18 (26)
No. of visits	Mean (SD)	12 (12)	16 (16)	12 (9)
Other home and community services†	n (%)	18 (30)	27 (39)	29 (42)
Aids and equipment†	n (%)	5 (8)	10 (14)	5 (7)
Respite care	n (%)	1 (2)	1 (1)	2 (3)
No. of d	Mean (SD)	1 (-)	1 (-)	3 (3)
Informal care	n (%)	30 (49)	33 (47)	36 (52)
No. of h per wk	Mean (SD)	20 (23)	25 (26)	17 (15)
Home modifications	n (%)	3 (5)	3 (4)	1 (1)
Patient employment loss	n (%)	0 (0)	1 (1)	2 (3)
Patient employment change	n (%)	2 (3)	2 (3)	1 (1)
Carer employment loss	n (%)	2 (3)	2 (3)	3 (4)
Carer employment gain	n (%)	0 (0)	0 (0)	1 (1)
Carer employment change	n (%)	9 (15)	8 (11)	5 (7)

n (%): number of participants (and percentage) who reported this event within the group. Mean (SD) is reported for those who reported the resource use. No significant differences were observed between groups.

*See Table S2.

†See Table S3.

using chained equations was used for missing values related to costs and QALYs. Imputed values were based on predictive mean matching with 5 nearest neighbors using age, time since stroke, modified Rankin Scale, and aphasia severity at baseline. Bootstrapping was used to test uncertainty around the differences in costs and outcomes between the intervention groups and usual care, with the cohorts resampled to have 1000 estimates of costs and benefits to determine the probability of cost-effectiveness. Analyses were conducted using Stata/SE 16.0 (StataCorp LLC).

RESULTS

In total, 201 (93.1%) participants completed the postintervention assessment and were included in the primary analysis (70 CIAT-Plus, 70 M-MAT, 61 Usual care). Demographic and clinical characteristics have been published elsewhere along with the results of the primary outcome analysis.[10] Mean age was 63 years, 68% presented with mild aphasia severity, and for 69% of participants their stroke had occurred more than 2 years before randomization. Few participants were in employment at baseline (3 in CIAT-Plus, 3 in M-MAT, and 7 in Usual care; P=0.346). Total costs were imputed for 1 participant in the M-MAT group who did not complete the cost questionnaire. QALYs were imputed for 8 participants (2 in CIAT-Plus, 3 in M-MAT, and 3 in Usual care) due to incomplete EQ-5D-3 L responses.

Before participation in the study, there were differences in the proportion of patients reporting hospital presentations and health services provided by specialists, with the M-MAT group having a lower proportion than other groups (Table S1). Between baseline and 12 weeks follow-up, there were no significant differences between groups in terms of resource utilization. The most commonly reported resource use items were outpatient rehabilitation and general practitioner visits

Table 3. Costs to 12 Weeks

	Usual care N=61	CIAT-Plus N=70	M-MAT N=69
	Mean AUD (SD)	Mean AUD (SD)	Mean AUD (SD)
Intervention	...	622 (87)	623 (105)
Emergency department	168 (410)	63 (206)	127 (306)
Hospital admissions	1349 (6065)	2448 (8899)	563 (3726)
Rehabilitation admissions	215 (1683)	318 (1882)	0 (0)
Outpatient rehabilitation	2732 (12 881)	2163 (4753)	2579 (8508)
General practitioner	147 (147)	107 (107)	130 (143)
Speech pathology	11 (49)	5 (39)	15 (96)
Social work	5 (33)	0 (0)	0 (0)
Psychologist	10 (47)	20 (115)	8 (48)
Psychiatrist	14 (75)	0 (0)	0 (0)
Neurologist*	92 (262)	91 (215)	23 (115)
Other health services†	617 (1122)	632 (1002)	352 (723)
Home rehabilitation	59 (222)	405 (1677)	257 (933)
Community based aphasia	180 (579)	190 (478)	69 (308)
Other home and community services‡	1217 (7215)	2315 (12 909)	713 (2551)
Aids and equipment‡	36 (222)	39 (189)	22 (181)
Private speech therapy	335 (927)	180 (533)	219 (716)
Respite care	1 (7)	1 (7)	5 (35)
Informal care	5707 (10 984)	6767 (12 369)	5274 (8051)
Home modifications	538 (4096)	12 (79)	7 (60)
Patient employment loss	0 (0)	169 (1417)	271 (1579)
Patient employment change	−100 (2014)	−16 (1449)	−439 (3644)
Carer employment loss	344 (1897)	320 (1995)	544 (2686)
Carer employment gain	0 (0)	0 (0)	−162 (1343)
Carer employment change	120 (6003)	628 (5226)	−96 (5108)

P values are presented in Table S7. Costs are expressed in 2020 Australian dollars (AUD). Negative values indicate a cost saving/offset. CIAT indicates constraint-induced aphasia therapy; and M-MAT, multimodality aphasia therapy.

*P<0.05 between usual care and M-MAT.

†See Table S2.

‡See Table S3.

Table 4. Costs and Outcomes to 12 Weeks

	Without multiple imputation			With multiple imputation		
	Usual Care	CIAT	M-MAT	Usual Care	CIAT	M-MAT
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
	N=61	N=70	N=69	N=61	N=70	N=70
Costs AUD, mean (SD)						
Health care cost	7726 (16 802)	8988 (17 142)	5089 (9900)	7726 (16 802)	8988 (17 142)	5135 (10 340)
Hospital	4464 (14 383)	4991 (11 853)	3269 (9158)	4464 (14 383)	4991 (11 853)	3360 (9628)
Community health care*	889 (1140)	1037 (1793)	743 (1489)	889 (1140)	1037 (1793)	749 (1500)
Societal cost	6071 (13 097)	7868 (14 677)	5392 (10 591)	6071 (13 097)	7868 (14 677)	5337 (10 535)
Total cost	13 797 (26 292)	17 478 (25 255)	11 113 (15 090)	13 797 (26 292)	17 478 (25 255)	11 113 (15 199)
Health-related quality of life						
Baseline	N=61	N=70	N=70			
No problems with, n (%)						
Mobility	32 (53)	41 (59)	44 (63)	…	…	…
Self-care	50 (82)	48 (69)	58 (83)	…	…	…
Usual activities	30 (49)	29 (41)	32 (46)	…	…	…
Pain/discomfort	31 (51)	41 (59)	46 (66)	…	…	…
Anxiety/depression	46 (75)	51 (73)	54 (77)	…	…	…
Visual analog scale, mean (SD)	77 (16)	74 (19)	76 (17)	…	…	…
Utility score, mean (SD)	0.78 (0.22)	0.78 (0.18)	0.81 (0.21)	…	…	…
12 wk	N=59	N=67	N=67			
No problems with, n (%)						
Mobility	28 (47)	38 (57)	43 (68)	…	…	…
Self care	47 (80)	56 (84)	51 (76)	…	…	…
Usual activities	31 (53)	36 (54)	31 (46)	…	…	…
Pain/discomfort	33 (56)	42 (63)	47 (70)	…	…	…
Anxiety/depression	45 (76)	54 (81)	52 (78)	…	…	…
Visual analog scale, mean (SD)	73 (16)	77 (15)	78 (16)	…	…	…
Utility score, mean (SD)	0.77 (0.22)	0.80 (0.21)	0.82 (0.21)	…	…	…
	N=59	N=67	N=67	N=61	N=70	N=70
QALYs to 12 wk, mean (SD)	0.19 (0.05)	0.20 (0.04)	0.20 (0.05)	0.19 (0.05)	0.20 (0.04)	0.20 (0.05)

No significant differences were observed between groups. *P* values are presented in Table S8. Costs are expressed in 2020 Australian dollars (AUD). CIAT indicates constraint-induced aphasia therapy; M-MAT, multimodality aphasia therapy; and QALY, quality-adjusted life year.

*Includes costs related to general practitioner, speech pathology, social work, psychologist, psychiatrist, neurologist, home rehabilitation, community-based aphasia, aids and equipment and private speech therapy.

(Table 2; Tables S2 through S5). Approximately one-third of patients reported outpatient rehabilitation (M-MAT, 32%; CIAT-Plus, 31%; Usual care, 25%), and of those who reported outpatient rehabilitation, the mean number of outpatient visits per group was the greatest in usual care (33 visits) and the lowest in CIAT-Plus (21 visits). Most patients reported general practitioner visits (M-MAT, 80%; Usual care, 79%; CIAT-Plus, 73%), and of those reporting general practitioner visits, there were 2 to 3 general practitioner visits per participant per group over the 12-week follow-up period. Approximately half of the participants reported receiving informal care, with between 17 and 25 hours on average per week reported per participant. There were participants who reported changes in employment for themselves (n=8) or their carer (n=30) during the study period.

The cost of neurologists was significantly different between usual care and M-MAT (Table 3). There were no other significant differences in costs between the groups. The greatest costs incurred were related to outpatient rehabilitation (over $2000 per participant per group) and informal care (over $5000 per participant per group). Apart from general practitioner visits, medians for different cost categories were $0 due to the majority of patients not incurring costs in that category.

Mean total costs were $13 797 for usual care, $17 478 for CIAT-Plus, and $11 113 for M-MAT (Table 4). Community health care costs were $889 for usual care, $1037 for CIAT-Plus, and $743 for M-MAT. There were no mean differences in costs, utilities, or QALYs between groups with or without multiple imputation. There were no median differences in costs, utilities, or QALYs between groups observed at baseline or 12 weeks after the end of intervention, and over 25% of participants in all groups had a utility score of 1.00 corresponding to perfect health (Table S6). There were large standard deviations in costs relative to the mean, due to the skewness of data (Figure S1). With only marginal differences between usual care and CIAT-Plus in QALYs (<0.01 QALYs), and large differences in total costs, the cost per QALY gained for CIAT-Plus was $597 802. When considering community health care costs only, the cost per QALY gained for CIAT-Plus was $24 116. M-MAT was cheaper on average than usual care (by $2684 for total costs and by $146 for community health care costs) with a QALY difference slightly >0.01.

In almost 80% of bootstrapped samples, M-MAT resulted in more QALYs and was cost saving when compared with usual care when total costs were considered (Figure 1). CIAT-Plus resulted in more QALYs but was more expensive when compared with usual care in over 50% of bootstrapped samples when total costs were considered. Similar results were observed when considering community health care costs (Figure 2).

DISCUSSION

We have performed an economic evaluation of COMPARE, which provides novel insights into the resource utilization and costs for people with chronic stroke and aphasia, and the costs and benefits of providing additional therapy for aphasia compared with usual care. The trial offered an important opportunity to capture the data from a population of survivors of stroke who are often excluded from participating in clinical trials.[22] Overall, it appeared that M-MAT was more favorable than CIAT-Plus when compared with usual care in terms of costs and QALYs. CIAT-Plus was likely to result in more QALYs, but at an additional cost when compared with usual care. These results were consistent with findings that M-MAT was favorable to CIAT-Plus for aphasia-related quality of life measured by the Stroke and Aphasia Quality of Life Scale-39 communication scale.[10] Although we did not detect statistically significant differences in costs related to hospital admissions, home and community services, and informal care, there were large differences in these costs between groups relative to other cost categories. We hypothesize that M-MAT generally had lower costs relative to other groups due to improvements in outcomes, which in turn decreases the utilization of health services. Several perspectives of costs were presented, and since serious adverse events in this study were unrelated to the intervention, a perspective of costs limited to community health care deemed most likely to be affected by the interventions was also presented. Depending on the perspective taken, CIAT-Plus may be considered to be cost-effective, even at a lower willingness-to-pay threshold of ≈AUD 28 000 per QALY gained.[23] In addition to the cost-effectiveness results, we provide novel

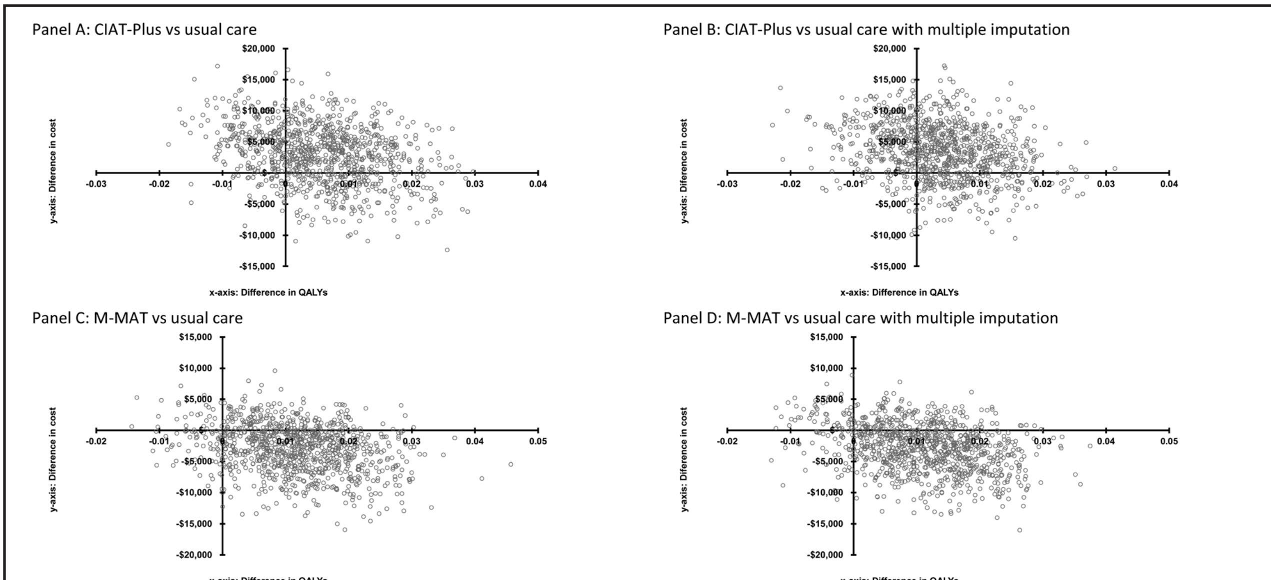

Figure 1 – Probability of cost effectiveness (total costs)

	Better outcomes and cost saving (dominant)	Better outcomes, more costly (cost effective n/N, %)	Poorer outcomes and more costly (inferior)	Poorer outcomes, less costly
Panel A: CIAT-Plus vs usual care	21.5%	51.8% (26/518, 5.0%)	23.1%	3.6%
Panel B: CIAT-Plus vs usual care with multiple imputation	18.1%	51.0% (20/510, 3.9%)	27.8%	3.1%
Panel C: M-MAT vs usual care	72.4%	19.1% (34/191, 17.8%)	2.8%	5.7%
Panel D: M-MAT vs usual care with multiple imputation	71.8%	17.7% (29/177, 16.4%)	5.8%	4.7%

Willingness to pay threshold used for cost effectiveness: $50,000 / QALY gained

Figure 1. Probability of cost-effectiveness (total costs).
CIAT indicates constraint-induced aphasia therapy; and M-MAT, multimodality aphasia therapy.

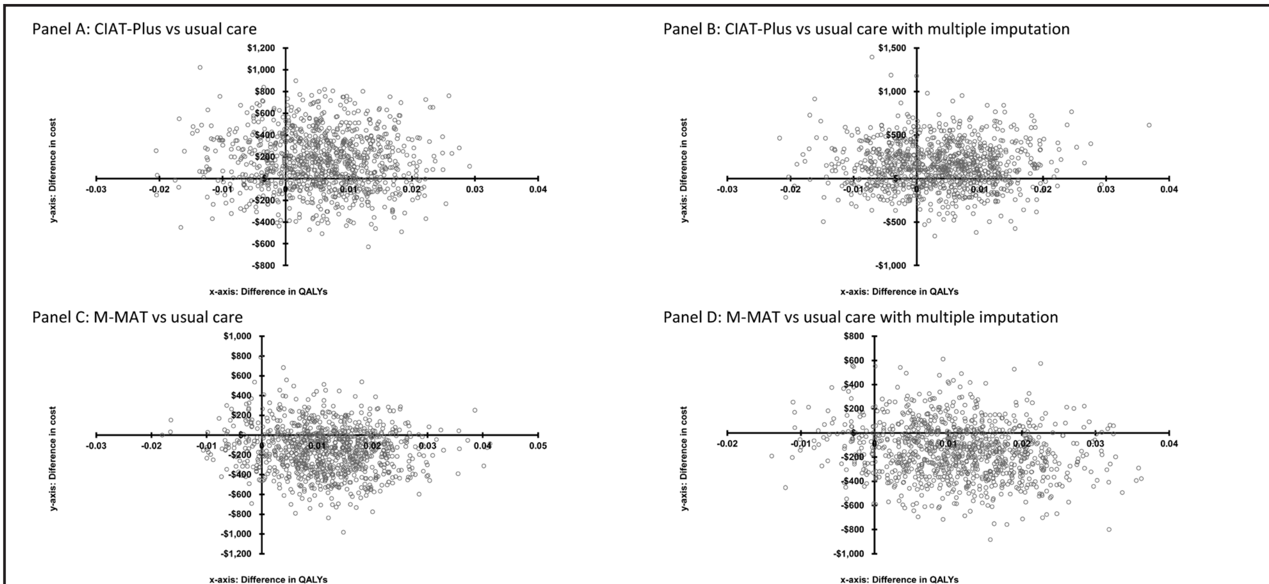

Figure 2 – Probability of cost effectiveness (community health care costs)

	Better outcomes and cost saving (dominant)	Better outcomes, more costly (cost effective n/N, %)	Poorer outcomes and more costly (inferior)	Poorer outcomes, less costly
Panel A: CIAT-Plus vs usual care	22.0%	53.8% (354/538, 65.8%)	17.2%	7.0%
Panel B: CIAT-Plus vs usual care with multiple imputation	19.2%	45.8% (308/458, 67.3%)	24.6%	10.4%
Panel C: M-MAT vs usual care	68.5%	22.2% (200/222, 90.1%)	3.2%	6.1%
Panel D: M-MAT vs usual care with multiple imputation	68.5%	22.1% (196/221, 88.7%)	3.3%	6.1%

Willingness to pay threshold used for cost effectiveness: $50,000 / QALY gained

Figure 2. Probability of cost-effectiveness (community health care costs).
CIAT indicates constraint-induced aphasia therapy; M-MAT, multimodality aphasia therapy; and QALY, quality-adjusted life year.

data on the utilization of health services and other economic impacts in this cohort.

Research on the cost-effectiveness of therapies for patients with aphasia after stroke is disparate.[4–7] There was some evidence from the 2 studies involving patients with chronic aphasia after stroke (conducted in the United Kingdom[5] and China[7]) that computerized word finding therapy was cost-effective for people with mild or moderate aphasia,[5] and that scalp acupuncture in addition to speech and language therapy was cost-effective when compared with speech and language therapy alone.[7] Comparisons to all of these studies are complicated by the the differences in the timing of therapy following stroke,[4,6] the perspective of costs (primarily costs related to government-funded services were included for the studies conducted in the United Kingdom),[5,6] and differences in health care and welfare systems between our study setting and China.[7] Our results reveal low use of rehabilitation, psychology, social work, and speech pathology services at baseline and study completion in a cohort with considerable health care needs. There is evidence that people with chronic aphasia after stroke have significant communication disability, and a high prevalence of mood disturbance, carer burden, loss of employment, and financial stress.[24–26] Low utilization of these health services likely reflects a lack of service availability/access rather than patient/carer need. This is supported by evidence that private practice (fee for service) speech pathology services were accessed twice as frequently as government services. The frequency of general practitioner visits would be considered to be above average over the course of a year (6–11 visits per year).[27] This highlights the potential role that general practitioners, as frequently accessed services, could play in health promotion and facilitation of reentry pathways to rehabilitation and allied health services for this vulnerable population.

The utility values in this study were derived using the EQ-5D-3L questionnaire, which has not yet been validated in people with aphasia after stroke. Therefore, QALYs estimated here may be limited by the lack of sensitivity of the EQ-5D-3L to quality of life in this cohort, and this may be the reason why the results related to utility values presented in this study did not align with other findings that treatment improved aphasia-related quality of life.[10] Of note, utility values were slightly greater on average in this study (>10 points) than the average reported in other studies.[28] This finding was surprising given that this cohort had chronic aphasia and aphasia has been associated with poor quality of life,[29–31] scoring the lowest in 1 study of 60 health conditions with 66 193 participants.[32] Additionally, this may be an artifact of recruitment bias, with participants in trials having a better quality of life in general than those who are unable to be recruited due to greater social isolation, and greater communication and physical disability. Further research to determine the sensitivity of the EQ-5D-3L when assessing the quality of life for people with aphasia is required.

In terms of intervention costs, only the time taken by therapists to provide the intervention was considered. Additional costs of training therapists to provide CIAT-Plus and M-MAT were not considered. However, these costs are low: only 2 hours of online therapy-specific training beyond additional training provided in research trial procedures. Therefore, the additional costs of scaling the provision of these therapies to treat more people with chronic aphasia would not be onerous.

Strengths of the study include the study design and the comprehensive data collected that provide novel insights on service utilization, and the costs for people with chronic stroke and aphasia. The data collection was feasible, with costs missing for only 1 participant included in the primary outcome analysis. Some caution in the interpretation of results is needed since the study was not powered for this economic evaluation, and there were some imbalances in health service use and quality of life between groups before participation in the study.

While the data collected were self-reported, the detailed trial costs questionnaire was prospectively completed supporting participant recall and providing additional scaffolding for communication challenges, with a relative/friend providing assistance where necessary. In future studies, it would be important to explore the feasibility of obtaining information about health service utilization through person-level data linkage to hospital or Medicare claims datasets. The data represents the Australian context which provides health care under a mixed Medicare and private insurance model. The costs may not be generalizable to other countries that do not offer the same range of services or reimbursement health insurance schemes.

Aphasia treatments resulted in better outcomes, with evidence that CIAT-Plus would be provided at an acceptable additional cost compared with usual care, while M-MAT may also result in cost savings compared with usual care. Along with the findings of the clinical evaluation, these data will be important for advocating for the implementation of these therapies for patients with chronic aphasia after stroke.

ARTICLE INFORMATION

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Supplemental Material

Tables S1–S8
Figure S1
CHEERS Checklist
Economic evaluation standardized questionnaire

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