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BMJ 2005;331;659-; originally published online 11 Aug 2005;
doi:10.1136/bmj.38520.451840.E0

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Papers

Randomised controlled trial of the Lidcombe programme of early stuttering intervention

Mark Jones, Mark Onslow, Ann Packman, Shelley Williams, Tika Ormond, Ilsa Schwarz, Val Gebski

Abstract

Objectives To evaluate the efficacy of the Lidcombe programme of early stuttering intervention by comparison to a control group.

Design A pragmatic, open plan, parallel group, randomised controlled trial with blinded outcome assessment.

Setting Two public speech clinics in New Zealand.

Participants Stuttering preschool children who presented to the speech clinics for treatment. Inclusion criteria were age 3-6 years and frequency of stuttering of at least 2% syllables stuttered. Exclusion criteria were onset of stuttering during the six months before recruitment and treatment for stuttering during the previous 12 months. 54 participants were randomised: 29 to the Lidcombe programme arm and 25 to the control arm. 12 of the participants were girls.

Intervention Lidcombe programme of early stuttering intervention.

Main outcome measures Frequency of stuttering was measured as the proportion of syllables stuttered, from audiotaped recordings of participants' conversational speech outside the clinic. Parents in both arms of the trial collected speech samples in three different speaking situations before randomisation and at three, six, and nine months after randomisation.

Results Analysis showed a highly significant difference ($P=0.003$) at nine months after randomisation. The mean proportion of syllables stuttered at nine months after randomisation was 1.5% (SD 1.4) for the treatment arm and 3.9% (SD 3.5) for the control arm, giving an effect size of 2.3% of syllables stuttered (95% confidence interval 0.8 to 3.9). This effect size was more than double the minimum clinically worthwhile difference specified in the trial protocol.

Conclusions The results provide evidence from a randomised controlled trial to support early intervention for stuttering. The Lidcombe programme is an efficacious treatment for stuttering in children of preschool age.

Introduction

Stuttering usually starts in the third and fourth years of life, after a period of apparently normal speech development. Around 5% of children begin to stutter.¹ Although the recovery rate without professional intervention is 74%,² the natural recovery rate of cases presenting to clinics has not been researched. To date, sex and family history of recovery are the major identified predictors of natural recovery. Girls are more likely to recover than boys, and children with a family history of recovery are more likely to

recover than those without such a history. Natural recovery does not seem to be related to severity of stuttering.³

The consensus now is that stuttering should be treated in the preschool years, primarily because it becomes less tractable as children get older. This is presumably because neural plasticity decreases with age. Also, it is not possible to know in advance whether an individual child will recover naturally. Early intervention in the preschool years is therefore essential. Once stuttering becomes chronic, communication can be severely impaired, with devastating social, emotional, educational, and vocational effects.^{4,5}

Several treatments for early stuttering are currently available,⁶ but only one, the Lidcombe programme, has been studied with phase I and II clinical trials.⁷ This programme is a behavioural treatment developed specifically for stuttering in children of preschool age (younger than 6). Considerable research into the programme has been conducted. Preliminary studies have produced positive outcomes, and stuttering has been shown to be no longer present, or remaining at very low levels, two to seven years after treatment.⁸⁻¹¹ The social validity and safety of the programme have been shown.¹² It does not seem to change children's behaviour other than speech or affect the attachment of children and parents or use of language.^{13,14} Duration of treatment and its predictors have been investigated in two independent file audits of preschool children attending specialist clinics, one in Australia¹⁵ and one in the United Kingdom.¹⁶

Although outcomes of the Lidcombe programme have consistently been shown to be positive,⁸⁻¹⁶ a randomised controlled trial would establish whether the effects of this intervention are significantly and clinically greater than those of natural recovery. We investigated the efficacy of the Lidcombe programme through comparison with a control group that received no formal treatment, in a pragmatic, open plan, parallel group, randomised controlled trial. Our primary hypothesis was that at nine months after randomisation, children in the treatment arm would be stuttering less than children in the control arm. The minimum worthwhile difference between the two arms was set at 1.0% of syllables stuttered.

Methods

The two treatment sites were the campus clinic at the University of Canterbury (Christchurch, New Zealand) and the clinic at the premises of the Stuttering Treatment and Research Trust (Auckland, New Zealand). The study population consisted of preschool children who presented to these speech clinics for treatment. We obtained written informed consent for all participants before randomisation. Inclusion criteria for the trial were an age at

recruitment of 3-6 years, stuttering as diagnosed by using standard procedures¹⁷ and at least 2% of syllables stuttered, and proficiency in English for children and parents. Exclusion criteria were treatment for stuttering during the previous 12 months and onset of stuttering in the six months before recruitment.

The children allocated to the Lidcombe programme arm received the treatment according to the programme manual. Throughout the programme, parents provide verbal contingencies for periods of stutter free speech and for moments of stuttering. This occurs in conversational exchanges with the child in the child's natural environment. The contingencies for stutter free speech are acknowledgment ("That was smooth"), praise ("That was good talking"), and request for self evaluation ("Were there any bumpy words then?"). The contingencies for unambiguous stuttering are acknowledgment ("That was a bit bumpy") and request for self correction ("Can you say that again?"). The programme is conducted under the guidance of a speech pathologist. During the first stage of the programme, a parent conducts the treatment for prescribed periods each day, and parent and child visit the speech pathologist once a week. The second stage starts when stuttering has been maintained at a frequency of less than 1.0% of syllables stuttered over three consecutive weeks inside and outside the clinic and is designed to maintain those low levels. Treatment is withdrawn, and the frequency of clinic visits decreases over a period of at least one year, providing stuttering remains at less than 1.0% of syllables stuttered. Details of the treatment are available in the guide for clinicians,⁷ and the treatment manual is available from the website of the Australian Stuttering Research Centre (www.fhs.usyd.edu.au/ASRC).

Compliance with treatment was established by the consulting clinicians who trained the parents in the procedures and with sample tape recordings of parents conducting the treatments with their children in everyday speaking environments. These recordings confirmed that parents were administering treatment in accordance with the treatment manual. The parents of children in the control arm were told that they would receive the Lidcombe programme at the end of the trial if it was shown to be efficacious and their child was still stuttering. They were told they could receive treatment during the trial at other clinics, providing it was not the Lidcombe programme.

We asked parents in both arms of the trial to use audiotape recorders to collect three samples of their child's conversational speech outside the clinic, before randomisation and then three, six, and nine months after randomisation. Parents had to record samples of their child speaking to a family member at home, to a non-family member at home, and to a non-family member away from home, ideally at a similar time of the day. Experienced speech pathology observers measured the proportion of syllables stuttered from each recording in real time by using an electronic, button press counter and timer. One observer assessed all recordings from the Auckland site and another from the Christchurch site. Intrajudge and interjudge reliability was assessed on a 5% sample of tape recordings from both sites.

The sample size required for the trial was calculated based on a two tailed test, 80% power, level of significance 5%, and a minimum clinically worthwhile difference at nine months after randomisation of 1.0% syllables stuttered. This is the minimum difference that a listener would be able to distinguish. On the basis of existing data¹⁵ we assumed an exponential distribution for the proportion of syllables stuttered. A sample size of 55 in each group was sufficient to detect the minimum clinically worthwhile difference. This sample size also accounted for a modest non-compliance rate of 10%.

We used SAS, version 8.02 for Windows (SAS Institute, Cary, NC, USA), and Stata/SE, version 8.0 for Windows (Stata Corporation, College Station, TX, USA), for our analyses. We used a two sample *t* test to perform the primary comparison of difference in the mean proportion of syllables stuttered at nine months after randomisation. The frequency of stuttering analysed for each child was an average of their nine month samples. We used least squares regression to estimate treatment effect in important subgroups and interaction terms in the regression models to test for heterogeneity. All analyses were by intention to treat. We used the last observation carried forward for two participants without follow-up tapes at nine months.

Treatment assignment and blinding

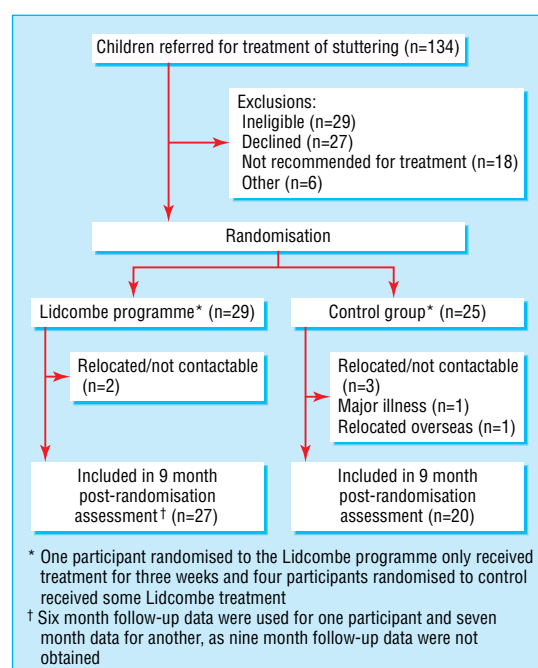
An independent central telephone randomisation service provided by the National Health and Medical Research Council (NHMRC) Clinical Trials Centre at the University of Sydney, Australia, assigned each eligible and consenting participant to the control group or treatment group. The allocation process entailed the treatment clinician contacting the council's clinical trials centre by telephone, fax, or email. Treatment assignment was conducted independently of the treating clinician. Dynamically balanced randomisation¹⁸ was used, with stratification by age, sex, severity of stuttering, treatment site, and family history of recovery from stuttering. Because blinding was not possible, observers blinded to treatment allocation assessed outcomes. All speech recordings were de-identified and masked to the allocated treatment.

Results

Because of difficulty with recruitment we decided to stop the trial before we had obtained 110 participants. Recruitment began in June 1999 and concluded in May 2003 with a total of 54 participants randomised, 31 at the University of Canterbury and 23 at the Stuttering Treatment and Research Trust.

Participant flow and follow-up

Seven (13%) of the 54 randomised participants did not complete the trial, and data after randomisation were not available, with all analyses being performed on 47 participants. Reasons for not completing the trial included major illness in the case of one child, and families being not contactable, mainly because of relocation. The figure shows the flow diagram of participants through the trial. Comparisons of the pre-randomisation characteristics for participants who were lost to follow-up with those remaining in the trial showed no statistically significant differences in severity, sex, treatment group assigned, or family history of recovery from stuttering (table 1). The difference in age reached significance, however, with the participants withdrawing being on average nine months older ($P = 0.015$, two sample *t* test). Two participants had data for analysis available for only six months and seven months, respectively, after randomisation. The median time from randomisation to final follow-up was 11 months in the control arm and nine months in the treatment arm. Five (9%) protocol violations occurred: four children in the control arm received some Lidcombe programme treatment and one child allocated to the intervention group received only three weeks of treatment (figure). Three participants allocated to the control arm received other treatment. Two participants received Easy-does-it,¹⁹ and one received some components of the Lidcombe programme on an ad hoc basis.



Flow of participants through the trial

Speech samples

We collected 517 speech samples for analysis. For each child at each of the measurement occasions in the trial, an average of 2.3 (range 1-6) speech samples were available for analysis. The speech samples obtained had a mean duration of 433 syllables (SD 12). We obtained intraclass correlations of $r=0.99$ for both intrajudge and interjudge reliability.

Analysis

Of the 54 participants recruited to the study, 29 were allocated to the treatment group and 25 to the control group. A comparison of pre-randomisation characteristics by treatment group shows the groups to be similar (table 2). Table 3 shows that severity of stuttering measured by proportion of syllables stuttered was similar in the two groups before randomisation. However, at nine months after randomisation, we noted a difference of 2.3% of syllables stuttered between the treatment group and the control group (95% confidence interval 0.8% to 3.9%, $P=0.003$). The results were similar after adjustment for treatment site, baseline severity, age, sex, and family history of recovery from stuttering.

Table 1 Comparison of baseline characteristics of participants lost to follow-up with participants remaining in study. Values are numbers (percentages) of subjects unless otherwise indicated

	Lost to follow up (n=7)	Remaining in study (n=47)	P value for difference (Fisher's exact test)
% syllables stuttered:			
2-5	3 (43)	15 (32)	0.7
>5	4 (57)	32 (68)	
Female	2 (29)	10 (21)	0.6
Family history of recovery	3 (43)	21 (45)	0.9
Treatment group:			
Lidcombe programme	2 (29)	27 (57)	0.2
Control	5 (71)	20 (43)	
Mean age in years (SD)	4.7 (0.7)	3.9 (0.6)	0.015 (two sample t test)

Table 2 Baseline characteristics of all participants. Values are numbers (%) of participants unless otherwise indicated

	Lidcombe programme (n=29)	Control (n=25)	Total (n=54)
Age in years:			
3-4	17 (59)	12 (48)	29 (54)
4-5	9 (31)	12 (48)	21 (39)
5-6	3 (10)	1 (4)	4 (7)
% syllables stuttered:			
2-5	9 (31)	9 (36)	18 (33)
>5	20 (69)	16 (64)	36 (67)
Female	7 (24)	5 (20)	12 (22)
Family history of recovery	13 (45)	11 (44)	24 (44)

We conducted an exploratory analysis of the proportion of children with less than 1.0% syllables stuttered at nine months after randomisation. The proportion was higher in the Lidcombe arm than in the control arm when adjusted for the baseline severity score in a logistic regression model (table 4). Table 5 shows estimates of effect size within subgroups stratified at baseline. The only test of heterogeneity to reach significance shows a larger effect of treatment for those children without a family history of recovery from stuttering than for children with a history. Effect sizes seemed to be consistent in all other important subgroups, with the possible exception of the treatment site. However, the control group doing better in Auckland than in Christchurch could explain this possible difference.

Discussion

After nine months, the reduction of stuttering in the Lidcombe programme group was significantly and clinically greater than natural recovery. The estimated effect size of 2.3% of syllables stuttered is more than double the minimum clinically worthwhile difference specified in the trial protocol. At nine months, the control group had reduced their frequency of stuttering by an average of 43%, presumably from a combination of natural recovery and the ad hoc treatment given to some of the participants. However, only 15% of children in the control arm attained a minimal level of stuttering as defined in the trial protocol. In contrast, the treatment group had reduced their stuttering by 77%, resulting in a mean frequency of 1.5% syllables stuttered. Most children in the Lidcombe programme group were still in the second stage of the programme at the nine month follow-up point.

Table 3 Severity of stuttering (% syllables stuttered) before randomisation and at nine months. Values are means with standard deviations unless otherwise indicated

	Lidcombe programme (n=27)	Control (n=20)	Difference in % syllables stuttered at nine months (95% CI, P value)
Before randomisation	6.4 (4.3)	6.8 (4.9)	2.3
At nine months	1.5 (1.4)	3.9 (3.5)	(0.8 to 3.9, $P=0.003$)

Table 4 Numbers (percentages) of participants achieving less than 1.0% of syllables stuttered at nine months

% syllables stuttered	Lidcombe programme (n=27)	Control group (n=20)	Odds ratio (95% CI)
<1.0	14 (52)	3 (15)	0.13
≥1.0	13 (48)	17 (85)	(0.03 to 0.63, $P=0.011$)

Table 5 Estimates of effect size within subgroups

Subgroup	No of participants (n=47)	Effect size as % syllables stuttered (95% CI)	P value for heterogeneity*
Treatment site:			
Auckland	22	1.1 (-0.6 to 2.8)	0.15
Christchurch	25	3.3 (0.9 to 5.8)	
Sex:			
Male	37	2.4 (0.6 to 4.2)	0.8
Female	10	2.0 (-1.6 to 5.5)	
Age in years:			
<4	28	2.4 (0.1 to 4.7)	0.9
≥4	19	2.3 (0.4 to 4.2)	
Family history of recovery:			
No	26	3.8 (1.5 to 6.2)	0.027
Yes	21	0.5 (-1.6 to 2.7)	
Baseline severity in % syllables stuttered:			
<5	19	2.1 (0.2 to 4.0)	0.6
≥5	28	2.7 (0.5 to 4.9)	

*Obtained from a test of interaction in a least squares regression model.

Study limitations

Our study has some limitations that should be acknowledged, but these are not sufficient to alter the main conclusion. The achieved sample size was only half that proposed, and seven of the 54 randomised participants did not complete the trial. These participants were older than the participants who did complete the trial, by an average of nine months. However, this difference in age is unlikely to have had an important effect on the results of the trial because the number of participants lost to follow-up is small, and there is no evidence to show that age is correlated with the proportion of syllables stuttered at nine months after randomisation for the participants who completed the trial.

Another limitation is that the post-randomisation period lasted nine months only. Ideally it should have been longer than this so that the children in the treatment group could have completed the second stage of the treatment programme and the children in the control group would have had more time to recover naturally. However, retaining a control group of stuttering children for longer than nine months is very difficult. Initially, follow-up was for 12 months, but this was reduced because parents of children allocated to the control group were unwilling to wait that long to receive treatment for their child.

Conclusions

Given its apparent efficacy, then, numerous reasons support implementing the Lidcombe programme in the preschool years. Although several children presenting at a clinic with early stuttering may recover without treatment, identifying these children in advance is not possible. Waiting for an extended period to see if natural recovery occurs is not acceptable because it seems that the Lidcombe programme is less efficacious once children move into the school age years.²⁰ In addition, delaying treatment until the school age years is not a viable option because of the negative social and cognitive consequences of stuttering at this age.²¹ If the disorder persists into the school age years a child is exposed to the unacceptable risk of experiencing the disabling effects of chronic and intractable stuttering throughout life.

We acknowledge the contribution of Marjorie Blakeley for treatment of participants at the University of Canterbury and Peta Forder for performing randomisation at the National Health and Medical Research Council Clinical Trials Centre at the University of Sydney.

Contributors: MJ, MO, AP, and VG contributed to initiation of the study, study design, conduct of the study, statistical analysis, and preparation of the

What is already known on this topic

Chronic stuttering in adulthood is intractable and has serious disabling effects

The consensus is that early intervention in the preschool years is necessary

Randomised controlled trial evidence for the efficacy of early stuttering intervention has not been obtained

What this study adds

The Lidcombe programme, an early intervention for stuttering, is significantly and clinically more efficacious than no formal programme in treating stuttering in preschool children

manuscript. SW and TO contributed to conduct of the study and preparation of the manuscript. IS contributed to initiation of the study and preparation of the manuscript. MO is the guarantor.

Funding: None.

Competing interests: None declared.

Ethical approval: Ethical approval was obtained from the human ethics committees at the University of Sydney and University of Canterbury prior to conducting the study.

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(Accepted 8 June 2005)

doi 10.1136/bmj.38520.451840.E0

Queensland Clinical Trials Centre, University of Queensland, Princess Alexandra Hospital, Woollongabba, Queensland 4102, Australia
Mark Jones *biostatistician*

Australian Stuttering Research Centre, University of Sydney, Lidcombe, NSW
2141, Australia

Mark Onslow *director*

Ann Packman *senior research officer*

Stuttering Treatment and Research Trust, Parnell, Auckland 1001, New Zealand
Shelley Williams *speech pathologist*

Department of Communication Disorders, University of Canterbury, Christchurch
8020, New Zealand

Tika Ormond *speech pathologist*

Department of Audiology and Speech Pathology, University of Tennessee,
Knoxville, Tennessee 37996, USA

Ilsa Schwarz *head of department*

National Health and Medical Research Council Clinical Trials Centre, University
of Sydney, Camperdown, NSW 2050, Australia

Val Gebski *head of biostatistics and research methodology*

Correspondence to: M Onslow m.onslow@fhs.usyd.edu.au