QUESTION

Should multi-faceted approach to physical therapy treatment vs. single focused therapy (i.e. strengthening only) be used for ambulant people with Friedreich ataxia?	
POPULATION:	ambulant people with Friedreich ataxia
INTERVENTION:	multi-faceted approach to physical therapy treatment
COMPARISON:	single focused therapy (i.e. strengthening only)
MAIN OUTCOMES:	Independence of ambulation; Independence of ambulation; Independence of ambulation; Independence of ambulation; Independence of ambulation; Independence of ambulation; Balance; Balance; Balance; Falls; Walking capacity; Quality of Life; Lower limb strength;

ASSESSMENT

Problem

Is the problem a priority?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ No ○ Probably no ○ Probably yes ● Yes ○ Varies ○ Don't know	Gait instability is the most frequently reported initial symptom in individuals with FRDA, occurring as the first symptom in 76 - 88% of individuals (Reetz et al, 2015). Mobility typically declines, with loss of mobility for individuals with onset <15 years of age typically 11.5 years after first symptom onset; 18.3 years for individuals with onset 15-24 years of age and 23.5 years for individuals with onset >24 years (Rummey et al, 2020).	The Friedreich's ataxia Clinical Management Guideline Patient and Parent Advisory Panel were interviewed on the consequences, urgency and priority of the topic. 1/7 indicated the consequences of the disturbance of strength, balance, mobility and reduction of falls were probably serious, 5/7 indicated serious, 1/7 indicated didn't know if serious. 1/7 indicated the consequences of the disturbance of strength, balance, mobility and reduction of falls were probably not urgent, 1/7 indicated probably urgent, 5/7 indicated urgent. 1/7 indicated the consequences of the disturbance of strength, balance, mobility and reduction of falls were probably a priority, 6/7 indicated priority. (Aug 2020).

Desirable Effects

How substantial are the desirable anticipated effects?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ Trivial ○ Small ● Moderate ○ Large ○ Varies ○ Don't know		In clinical practice, positive short-term outcomes from multi-faceted rehabilitation programs that are provided by physiotherapists specialising in the management of neurological conditions have been seen. However, long term outcomes appear variable and are based on continued participation in physical activities or exercise and rate of decline for the individual at that point in time.

Outcomes	№ of	Certainty of	Relative	Anticipated absolute effects* (95% CI)

	participants (studies) Follow-up	the evidence (GRADE)	effect (95% CI)	Risk with single focused therapy (i.e. strengthening only)	Risk difference with multi-faceted approach to physical therapy treatment
Independence of ambulation assessed with: Functional Ambulation Category	0 (1 observational study)[1]	⨁◯◯◯ Very low[a,b,c]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the FAC pre- and post- treatment (p=0.016).	
Independence of ambulation assessed with: Hoffer Ambulation Scale	0 (1 observational study)[1]	⨁◯◯◯ Very low[a,b,c]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the Hoffer Ambulation scale pre- and post-treatment (p<0.05).	
Independence of ambulation assessed with: Barthel Index	0 (2 RCTs)[1,2]	⨁◯◯◯ Very low[a,b,d,e]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the Barthel Index pre- (mean 46.8, SD 8.8) and post- (mean 54.2, SD 10.3) treatment (p<0.001). (Dogan-Aslan et al 2018). 18 people with spinocerebellar ataxia type 7 (SCA7) were randomised to intensive training (2hr sessions, 5x/week for 24 weeks, n=5), moderate training (2hr sessions, 3x/week for 24 weeks, n=6) and non-training groups (n=7). A two-way, repeated-measure ANOVA identified no significant improvement in	

				the Barthel Index. (Tercero-Perez et al 2019).
Independence of ambulation assessed with: Functional Independence Measure	0 (2 RCTs)[3]	⊕⊕⊕◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Rehabilitation effectiveness was analysed using independent sample t-tests to compare change from baseline and six-week visit between groups. No significant difference was found in the FIM, between the groups from baseline to six-week visit. There was no significant between-group difference in the FIM motor domain, however there was a significant within-group increased for the intervention group. (Milne et al 2018).
Independence of ambulation assessed with: Friedreich Ataxia Rating Scale	0 (1 RCT)[3]	-	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 97.3, SD 22.2) and post-HEP (mean 94.0, SD 23.2) were found in the FARS (p=0.017). (Milne et al 2018).
Independence of ambulation assessed with: Scale for the Assessment and Rating of Ataxia	0 (3 RCTs)[2,4,5]	⊕⊕◯◯ Low[a,b,f,g]	-	30 people with spinocerebellar ataxia type 2 were randomised to either intensive exercise rehabilitation (4hr/day, 5x/week, 12 weeks, n=15) or no rehabilitation (n=15). Repeated-measures ANOVA of group x time were used to determine changes in outcome

				measures - there were no significant group x time interactions in the SARA. (Velazquez-Perez et al 2019). 18 people with spinocerebellar ataxia type 7 (SCA7) were randomised to intensive training (2hr sessions, 5x/week for 24 weeks, n=5), moderate training (2hr sessions, 3x/week for 24 weeks, n=6) and non-training groups (n=7). A two-way, repeated-measure ANOVA identified a significant reduction in SARA score (p<0.05) in the moderate (0.5 points) and intensive (1.4 points) training groups compared to non-training group. (Tercero-Perez et al 2019). 38 people with spinocerebellar ataxia type 2 were randomised to treatment (6 hrs/5 days/week, 24 weeks, n=19) or no treatment (n=19). Repeated-measure ANOVA was used to compare treatment and control groups - the treatment group showed a significant decrease in the SARA score at 24 weeks compared to control group. (Rodriguez-Diaz et al 2018).
Balance assessed with: Berg Balance Scale	0 (1 RCT)[3]	⊕⊕⊕◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline and post-rehab were found in the Berg Balance Scale (p=0.039). as well as between baseline and post-HEP (p=0.026) for non-ambulant participants. (Milne et al 2018).
Balance assessed with: Friedreich	0 (1 RCT)[3]	⊕⊕⊕◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme

Ataxia Rating Scale				immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 97.3, SD 22.2) and post-HEP (mean 94.0, SD 23.2) were found in the FARS (p=0.017). (Milne et al 2018).	
Balance assessed with: Scale for the Assessment and Rating of Ataxia	0 (2 RCTs)[4,5]	⨁⨁◯◯ Low[a,b,h,i]	-	30 people with spinocerebellar ataxia type 2 were randomised to either intensive exercise rehabilitation (4hr/day, 5x/week, 12 weeks, n=15) or no rehabilitation (n=15). Repeated-measures ANOVA of group x time were used to determine changes in outcome measures - there were no significant group x time interactions in the SARA. (Velazquez-Perez et al 2019). 38 people with spinocerebellar ataxia type 2 were randomised to treatment (6 hrs/5 days/week, 24 weeks, n=19) or no treatment (n=19). Repeated-measure ANOVA was used to compare treatment and control groups - the treatment group showed a significant decrease in the SARA score at 24 weeks compared to control group. (Rodriguez-Diaz et al 2018).	
Falls - not measured	-	-	-	-	-
Walking capacity - not measured	-	-	-	-	-
Quality of Life assessed with: Friedreich Ataxia Impact	0 (1 RCT)[3]	⨁⨁⨁◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control	

Scale				group). The rehabilitation was followed by a six-week home exercise programme. Rehabilitation effectiveness was analysed using independent sample t-tests to compare change from baseline and six-week visit between groups. There was a significant between-group difference in the FAIS body movement scale (p=0.003). Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 35.2 SD 21.9) and post-rehab (mean 27.7, SD 19.7) were found in the FAIS body movement scale (p=0.009) and in the FAIS lower limb (mean 65.2, SD 28.5 to mean 48.5, SD 31.5, p=0.033) and upper limb subscales (mean 29.3, SD 23.7 to mean 25.2, SD 20.4, p=0.044). (Milne et al 2018).	
Lower limb strength - not measured	-	-	-	-	-

1. Dogan-Aslan M., Buyukvural-Sen S.,Nakipoglu-Yuzer G.F.,Ozgirgin N. Demographic and clinical features and rehabilitation outcomes of patients with Friedreich ataxia: A retrospective study. Turkish Journal of Physical Medicine and Rehabilitation; 2018.
2. Tercero-Pérez, K.,Cortés,H.,Torres-Ramos,Y.,Rodríguez-Labrada,R.,Cerecedo-Zapata,C. M.,Hernández-Hernández,O.,Pérez-González,N.,González-Piña,R.,Leyva-García,N.,Cisneros,B.,Velázquez-Pérez,L.,& Magaña,J. J. Effects of Physical Rehabilitation in Patients with Spinocerebellar Ataxia Type 7. Cerebellum; 2019.
3. Milne S.C., Corben L.A.,Roberts M.,et al. Can rehabilitation improve the health and well-being in Friedreich's ataxia: a randomized controlled trial?. Clinical Rehabilitation; 2018.
4. Rodriguez-Diaz J.C., Velazquez-Perez L. Rodriguez Labrada R. et al. Neurorehabilitation therapy in spinocerebellar ataxia type 2: A 24-week, rater-blinded, randomized, controlled trial. Movement Disorders; 2018.
5. Velázquez-Pérez, L. Rodríguez-Diaz J. C. Rodríguez-Labrada R. Medrano-Montero J. Aguilera Cruz A. B. Reynaldo-Cejas L. Góngora-Marrero M. Estupiñán-Rodríguez A. Vázquez-Mojena Y. & Torres-Vega R. Neurorehabilitation Improves the Motor Features in Prodromal SCA2: A Randomized, Controlled Trial. Movement Disorders; 2019.

a. Small sample size.

b. Confidence intervals not reported.
c. No control group.
d. 18 participants with a diagnosis of spinocerebellar ataxia type 7, 37 participants with a diagnosis of FRDA.
e. One study a retrospective observational study.
f. Outcome measure no specific to measuring independence of ambulation.
g. Participants with a diagnosis of spinocerebellar type 2 or type 7 (no FRDA).
h. All participants with a diagnosis of spinocerebellar ataxia type II.
i. Outcome measure not specific to evaluating balance.

Undesirable Effects

How substantial are the undesirable anticipated effects?

JUDGEMENT

○ Large
○ Moderate
○ Small
● Trivial
○ Varies
○ Don't know

RESEARCH EVIDENCE

Outcomes	№ of participants (studies) Follow-up	Certainty of the evidence (GRADE)	Relative effect (95% CI)	Anticipated absolute effects* (95% CI)	
				Risk with single focused therapy (i.e. strengthening only)	Risk difference with multi-faceted approach to physical therapy treatment
Independence of ambulation assessed with: Functional Ambulation Category	0 (1 observational study)[1]	⨁◯◯◯ Very low[a,b,c]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the FAC pre- and post- treatment (p=0.016).	
Independence of ambulation assessed with: Hoffer Ambulation Scale	0 (1 observational study)[1]	⨁◯◯◯ Very low[a,b,c]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the	

ADDITIONAL CONSIDERATIONS

Fatigue may be a negative outcome from participating in a multi-faceted rehabilitation program, but this is no different to working on a single-focused approach to rehabilitation and should be managed at an individual level. In clinical practice, immediate/initial reports of fatigue don't appear have a lasting effect.

				Hoffer Ambulation scale pre- and post-treatment (p<0.05).
Independence of ambulation assessed with: Barthel Index	0 (2 RCTs)[1,2]	⨁◯◯◯ Very low[a,b,d,e]	-	37 people with Friedriech ataxia who received inpatient rehabilitation between Jan 2000 and Dec 2012 were included in this retrospective study. Mean rehabilitation duration was 4.7 weeks (SD 1.4 weeks). At the end of the rehabilitation program, a statistically significant difference was found in the Barthel Index pre- (mean 46.8, SD 8.8) and post- (mean 54.2, SD 10.3) treatment (p<0.001). (Dogan-Aslan et al 2018). 18 people with spinocerebellar ataxia type 7 (SCA7) were randomised to intensive training (2hr sessions, 5x/week for 24 weeks, n=5), moderate training (2hr sessions, 3x/week for 24 weeks, n=6) and non-training groups (n=7). A two-way, repeated-measure ANOVA identified no significant improvement in the Barthel Index. (Tercero-Perez et al 2019).
Independence of ambulation assessed with: Functional Independence Measure	0 (2 RCTs)[3]	⨁⨁⨁◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Rehabilitation effectiveness was analysed using independent sample t-tests to compare change from baseline and six-week visit between groups. No significant difference was found in the FIM, between the groups from baseline to six-week visit. There was no significant between-group difference in the FIM motor domain, however there was a significant within-group increased for the intervention group. (Milne et al 2018).
Independence of ambulation	0 (1 RCT)[3]	-	-	19 participants with Friedreich ataxia were randomised to a six-week

assessed with: Friedreich Ataxia Rating Scale				outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 97.3, SD 22.2) and post-HEP (mean 94.0, SD 23.2) were found in the FARS (p=0.017). (Milne et al 2018).	
Independence of ambulation assessed with: Scale for the Assessment and Rating of Ataxia	0 (3 RCTs)[2,4,5]	⊕⊕◯◯ Low[a,b,f,g]	-	30 people with spinocerebellar ataxia type 2 were randomised to either intensive exercise rehabilitation (4hr/day, 5x/week, 12 weeks, n=15) or no rehabilitation (n=15). Repeated-measures ANOVA of group x time were used to determine changes in outcome measures - there were no significant group x time interactions in the SARA. (Velazquez-Perez et al 2019). 18 people with spinocerebellar ataxia type 7 (SCA7) were randomised to intensive training (2hr sessions, 5x/week for 24 weeks, n=5), moderate training (2hr sessions, 3x/week for 24 weeks, n=6) and non-training groups (n=7). A two-way, repeated-measure ANOVA identified a significant reduction in SARA score (p<0.05) in the moderate (0.5 points) and intensive (1.4 points) training groups compared to non-training group. (Tercero-Perez et al 2019). 38 people with spinocerebellar ataxia type 2 were randomised to treatment (6 hrs/5 days/week, 24 weeks, n=19) or no treatment (n=19). Repeated-measure ANOVA was used to compare treatment and control groups - the treatment group showed a significant decrease in the SARA score at 24 weeks compared to control group. (Rodriguez-Diaz et al 2018).	

Balance assessed with: Berg Balance Scale	0 (1 RCT)[3]	⊕⊕⊕◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline and post-rehab were found in the Berg Balance Scale (*p*=0.039). as well as between baseline and post-HEP (*p*=0.026) for non-ambulant participants. (Milne et al 2018).
Balance assessed with: Friedreich Ataxia Rating Scale	0 (1 RCT)[3]	⊕⊕⊕◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 97.3, SD 22.2) and post-HEP (mean 94.0, SD 23.2) were found in the FARS (*p*=0.017). (Milne et al 2018).
Balance assessed with: Scale for the Assessment and Rating of Ataxia	0 (2 RCTs)[4,5]	⊕⊕◯◯ Low[a,b,h,i]	-	30 people with spinocerebellar ataxia type 2 were randomised to either intensive exercise rehabilitation (4hr/day, 5x/week, 12 weeks, n=15) or no rehabilitation (n=15). Repeated-measures ANOVA of group x time were used to determine changes in outcome measures - there were no significant group x time interactions in the SARA. (Velazquez-Perez et al 2019). 38 people with spinocerebellar ataxia type 2 were randomised to treatment (6 hrs/5

				days/week, 24 weeks, n=19) or no treatment (n=19). Repeated-measure ANOVA was used to compare treatment and control groups - the treatment group showed a significant decrease in the SARA score at 24 weeks compared to control group. (Rodriguez-Diaz et al 2018).	
Falls - not measured	-	-	-	-	-
Walking capacity - not measured	-	-	-	-	-
Quality of Life assessed with: Friedreich Ataxia Impact Scale	0 (1 RCT)[3]	⨁⨁⨁◯ Moderate[a]	-	19 participants with Friedreich ataxia were randomised to a six-week outpatient rehabilitation programme immediately (intervention group) or after a six-week delayed-start (control group). The rehabilitation was followed by a six-week home exercise programme. Rehabilitation effectiveness was analysed using independent sample t-tests to compare change from baseline and six-week visit between groups. There was a significant between-group difference in the FAIS body movement scale (p=0.003). Paired t-tests were used to determine change in baseline and immediately post-rehab, and baseline and immedidately after post-home exercise programme (HEP). Significant improvements between baseline (mean 35.2 SD 21.9) and post-rehab (mean 27.7, SD 19.7) were found in the FAIS body movement scale (p=0.009) and in the FAIS lower limb (mean 65.2, SD 28.5 to mean 48.5, SD 31.5, p=0.033) and upper limb subscales (mean 29.3, SD 23.7 to mean 25.2, SD 20.4, p=0.044). (Milne et al 2018).	
Lower limb strength - not	-	-	-	-	-

measured						

1. Dogan-Aslan M., Buyukvural-Sen S.,Nakipoglu-Yuzer G.F.,Ozgirgin N. Demographic and clinical features and rehabilitation outcomes of patients with Friedreich ataxia: A retrospective study. Turkish Journal of Physical Medicine and Rehabilitation; 2018.
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5. Velázquez-Pérez, L. Rodríguez-Diaz J. C. Rodríguez-Labrada R. Medrano-Montero J. Aguilera Cruz A. B. Reynaldo-Cejas L. Góngora-Marrero M. Estupiñán-Rodríguez A. Vázquez-Mojena Y. & Torres-Vega R. Neurorehabilitation Improves the Motor Features in Prodromal SCA2: A Randomized, Controlled Trial. Movement Disorders; 2019.

a. Small sample size.
b. Confidence intervals not reported.
c. No control group.
d. 18 participants with a diagnosis of spinocerebellar ataxia type 7, 37 participants with a diagnosis of FRDA.
e. One study a retrospective observational study.
f. Outcome measure no specific to measuring independence of ambulation.
g. Participants with a diagnosis of spinocerebellar type 2 or type 7 (no FRDA).
h. All participants with a diagnosis of spinocerebellar ataxia type II.
i. Outcome measure not specific to evaluating balance.

Certainty of evidence

What is the overall certainty of the evidence of effects?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS

○ Very low ○ Low ● Moderate ○ High ○ No included studies	Very low to moderate certainty of evidence as per the evidence profile table.	This criterion is judged as moderate due to the consistency of evidence supporting multi-faceted rehabilitation programs.

Values

Is there important uncertainty about or variability in how much people value the main outcomes?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ Important uncertainty or variability ○ Possibly important uncertainty or variability ○ Probably no important uncertainty or variability ● No important uncertainty or variability		

Outcomes	Importance	Certainty of the evidence (GRADE)
Independence of ambulation assessed with: Functional Ambulation Category	IMPORTANT[a]	⨁◯◯◯ VERY LOW[b,c,d]
Independence of ambulation assessed with: Hoffer Ambulation Scale	IMPORTANT[a]	⨁◯◯◯ VERY LOW[b,c,d]
Independence of ambulation assessed with: Barthel Index	IMPORTANT[a]	⨁◯◯◯ VERY LOW[b,c,e,f]
Independence of ambulation assessed with: Functional Independence Measure	IMPORTANT[a]	⨁⨁⨁◯ MODERATE[b]
Independence of ambulation assessed with: Friedreich Ataxia Rating Scale	IMPORTANT[a]	-
Independence of ambulation assessed with: Scale for the Assessment and Rating of Ataxia	IMPORTANT[a]	⨁⨁◯◯ LOW[b,c,g,h]
Balance	IMPORTANT[i]	⨁⨁⨁◯

assessed with: Berg Balance Scale		MODERATE[b]
Balance assessed with: Friedreich Ataxia Rating Scale	IMPORTANT[i]	⊕⊕⊕◯ MODERATE[b]
Balance assessed with: Scale for the Assessment and Rating of Ataxia	IMPORTANT[i]	⊕⊕◯◯ LOW[b,c,j,k]
Falls - not measured	CRITICAL[l]	-
Walking capacity - not measured	IMPORTANT[m]	-
Quality of Life assessed with: Friedreich Ataxia Impact Scale	CRITICAL[n]	⊕⊕⊕◯ MODERATE[b]
Lower limb strength - not measured	IMPORTANT[o]	-

a. Identified as critical (1/6), important (3/6) and low importance (2/6) by people with FA and critical by expert authors on the topic
b. Small sample size.
c. Confidence intervals not reported.
d. No control group.
e. 18 participants with a diagnosis of spinocerebellar ataxia type 7, 37 participants with a diagnosis of FRDA.
f. One study a retrospective observational study.
g. Outcome measure no specific to measuring independence of ambulation.
h. Participants with a diagnosis of spinocerebellar type 2 or type 7 (no FRDA).
i. Identified as critical (2/5) and important (3/5) by people with FA and important by expert authors on the topic.
j. All participants with a diagnosis of spinocerebellar ataxia type II.
k. Outcome measure not specific to evaluating balance.
l. Identified as critical (3/5) and important (2/5) by people with FA and important by expert authors on the topic
m. Identified as critical (2/6), important (3/6) and low importance (1/6) by people with FA and important by expert authors on the topic
n. Identified as critical (3/6) and important (3/6) by people with FA and critical by expert authors on the topic
o. Identified as critical (1/6) and important (5/6) by people with FA and important by expert authors on the topic

Balance of effects

Does the balance between desirable and undesirable effects favor the intervention or the comparison?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ Favors the comparison ○ Probably favors the comparison ○ Does not favor either the intervention or the comparison ○ Probably favors the intervention ● Favors the intervention ○ Varies ○ Don't know		A survey designed to systematically collect expert-based opinions from clinicians involved in developing the recommendations for this topic and providing clinical care for individuals with Friedreich ataxia, was conducted. Clinical experts from Australia, Europe, UK, South America, Canada and the USA were asked to consider the harms/benefits of **a multi-faceted approach to physical therapy treatment as a management strategy for ambulant individuals**. Reflecting on the impact of **a multi-faceted approach to physical therapy treatment** on Independence of ambulation, 100% (2/2) clinical experts reported a benefit (large, moderate or small), 0% (0/2) reported no effect and, 0% (0/2) reported observing a harm (large, moderate or small). Reflecting on the impact on Balance, 100% (2/2) clinical experts reported a benefit. Reflecting on the impact on Falls, 100% (2/2) clinical experts reported a benefit. Reflecting on the impact on Walking capacity, 100% (2/2) clinical experts reported a benefit. Reflecting on the impact on Quality of life, 100% (2/2) clinical experts reported a benefit. Reflecting on the impact on Lower Limb Strength, 100% (2/2) clinical experts reported a benefit.

Acceptability

Is the intervention acceptable to key stakeholders?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ No ○ Probably no ○ Probably yes ○ Yes ○ Varies ○ Don't know	No published evidence.	

SUMMARY OF JUDGEMENTS

	JUDGEMENT						
PROBLEM	No	Probably no	Probably yes	**Yes**		Varies	Don't know
DESIRABLE EFFECTS	Trivial	Small	**Moderate**	Large		Varies	Don't know
UNDESIRABLE EFFECTS	Large	Moderate	Small	**Trivial**		Varies	Don't know
CERTAINTY OF EVIDENCE	Very low	Low	**Moderate**	High			No included studies
VALUES	Important uncertainty or variability	Possibly important uncertainty or variability	Probably no important uncertainty or variability	**No important uncertainty or variability**			
BALANCE OF EFFECTS	Favors the comparison	Probably favors the comparison	Does not favor either the intervention or the comparison	Probably favors the intervention	**Favors the intervention**	Varies	Don't know
ACCEPTABILITY	No	Probably no	Probably yes	Yes		Varies	Don't know

TYPE OF RECOMMENDATION

Strong recommendation against the intervention	Conditional recommendation against the intervention	Conditional recommendation for either the intervention or the comparison	Conditional recommendation for the intervention	**Strong recommendation for the intervention**
○	○	○	○	●

CONCLUSIONS

Recommendation

For ambulant people with Friedreich ataxia, we recommend a multi-faceted rehabilitation approach (targeting multiple areas of impairment) over a single focused rehabilitation approach.

Justification

This recommendation is based on moderate evidence in favour of a multi-faceted rehabilitation approach and positive short-term outcomes seen in clinical practice.

Subgroup considerations

This recommendation is for individuals with Friedreich ataxia who are ambulant.

Research priorities

Randomised controlled trials examining short- and long-term outcomes from participating in a multi-faceted rehabilitation program are warranted to ensure equitable access and to provide further clarity on the benefits of rehabilitation for individuals with FRDA. Appropriate dosage of these programs also requires further exploration.

References

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