QUESTION

Should intensive (upper limb) rehabilitation vs. no rehabilitation be used for all people with Friedreich ataxia?	
POPULATION:	all people with Friedreich ataxia
INTERVENTION:	intensive (upper limb) rehabilitation
COMPARISON:	no rehabilitation
MAIN OUTCOMES:	Activities of daily living; Activities of daily living; Quality of life; Quality of life; Quality of life; Neurological function; Neurological function;
SETTING:	
PERSPECTIVE:	
BACKGROUND:	
CONFLICT OF INTERESTS:	

ASSESSMENT

Problem

Is the problem a priority?

JUDGEMENT	RESEARCH EVIDENCE	ADDITIONAL CONSIDERATIONS
○ No ○ Probably no ○ Probably yes ● Yes ○ Varies ○ Don't know		The Friedreich's ataxia Clinical Management Guideline Patient and Parent Advisory Panel were interviewed on the consequences, urgency and priority of the topic. 8/8 indicated upper limb dysfunction was serious. 1/7 indicated upper limb dysfuction was not urgent; 1/7 indicated probably not urgent; 1/7 indicated probably urgent; 4/7 indicated urgent. 1/7 indicated upper limb dysfunction was probably not a priority, 3/7 indicated probably a priority, 3/7 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

Outcomes	№ of participants (studies) Follow-up	Certainty of the evidence (GRADE)	Relative effect (95% CI)	Anticipated absolute effects* (95% CI)	
				Risk with no rehabilitation	Risk difference with intensive (upper limb) rehabilitation
Activities of daily living assessed with: ABILHAND scale	0 (1 RCT)[1]	⨁◯◯◯ Very low[a,b,c,d,e]	-	26 patients with progressive multiple sclerosis were randomised into active (ATG, n=13) or passive treatment groups (PTG, n=13). Each group underwent 36 1-hr treatment sessions, twice a week. Paired t-tests were used to determine changes between baseline and post-treatment for the ABILHAND scale. No significant changes were found in the ATG group. Significant worsening was found in the PTG group (p=0.02).	
Activities of daily living assessed with: Barthel index	0 (1 RCT)[2]	⨁◯◯◯ Very low[a,b,c,d,e]	-	19 patients with multiple sclerosis were randomly divided into 2 groups: exercise (n=10) and no exercise (n=9). The exercise group exercised with a physiotherapy 2 days/week, 60 min/session and performed independent home exercise 3 days/week for 4 weeks, >20mins/session. The no exercise group performed no exercises. A 2-way mixed-model repeated-measures ANOVA (time x intervention) demonstrated no statistically significant interaction in the Barthel Index.	
Quality of life assessed with: Modified fatigue impact scale	0 (3 RCTs)[1,3,4]	⨁◯◯◯ Very low[a,c,e]	-	26 patients with progressive multiple sclerosis were randomised into active (ATG, n=13) or passive treatment groups (PTG, n=13). Each group underwent 36 1-hr treatment sessions, twice a week. The interaction treatment group x time in RM ANCOVA showed a significant improvement, post-treatment versus baseline, in the ATG versus the PTG, in terms of MFIS total (p = 0.03) and physical (p = 0.01) scores. (Boffa et al 2019). 19 individuals with multiple sclerosis were divided into 2 groups: exercise (n=10, controlled group 2 days/week, 60 min/session with independent home exercise 3 days/week, >=20min/session), no exercise (n=9). A two-way mixed-model repeated-measures ANOVA identified a statistically significant group by time interaction on the MFIS scores for the exercise non-ambulatory subjects: physical (p=0.009), psychosocial (p=0.018), total (p=0.0008) scores. (Grubic Kezele et al 2019). 22 people with multiple sclerosis were randomly assigned to Group A (8 week rehabilitation followed by 8 weeks of no intervention), or Group B (same treatment in reverse order). An analysis of covariance showed a statistically significant treatment effect in the	

				MFIS (P = 0.05) without any carryover effect (P = 0.63). MFIS was lower after the treatment (T) compared to the waiting list (WL). The combined differences for Groups A and B between WL–T periods for MFIS was [median and interquartile range (Q1–Q3)] 5.2 (10.7) points. (Gervasoni et al 2019).
Quality of life assessed with: Visual Analogue Scale for pain	0 (1 RCT)[2]	⨁◯◯◯ Very low[a,b,c,d,e]	-	19 patients with multiple sclerosis were randomly divided into 2 groups: exercise (n=10) and no exercise (n=9). The exercise group exercised with a physiotherapy 2 days/week, 60 min/session and performed independent home exercise 3 days/week for 4 weeks, >20mins/session. The no exercise group performed no exercises. A 2-way mixed-model repeated-measures ANOVA (time x intervention) demonstrated a statistically significant group-by-time interaction in the visual analogue scale for pain only in non-ambulatory (*p*=0.049) individuals and not in ambulatory individuals (*p*=0.159).
Quality of life assessed with: SF-36	0 (1 RCT)[4]	⨁◯◯◯ Very low[a,b,c,d,e]	-	19 individuals with multiple sclerosis were divided into 2 groups: exercise (n=10, controlled group 2 days/week, 60 min/session with independent home exercise 3 days/week, >=20min/session), no exercise (n=9). A two-way mixed-model repeated-measures ANOVA identified a statistically significant group by time interaction on the physical functioning (SF-36) (p=0.014) and general health (SF-36) scores (*p*=0.042) in ambulatory subjects.
Neurological function assessed with: 9HPT	0 (5 RCTs)[1,3,5,6,7]	⨁◯◯◯ Very low[a,f,g]	-	22 people with multiple sclerosis were randomly assigned to Group A (8 week rehabilitation followed by 8 weeks of no intervention), or Group B (same treatment in reverse order). An analysis of covariance showed no statistically significant treatment effect in the 9HPT (*p*=0.63) with no carryover effect (*p*=0.67) (Gervasoni et al 2019). 30 people with multiple sclerosis were randomised into 2 groups of n=15, undergoing 20 1hr sessions, 3x/week for 2 months. The treatment group received active motor rehabilitation treatment while the control group received passive mobilisation of the shoulder, elbow, wrist and fingers. Factorial ANOVA with repeated measures (time x group) identified a statistically significant improvement as effect of time was found in time required to complete 9HPT (*p*<0.000001). However, there was no significant time x group interaction found in the 9HPT (*p*=0.98). (Bonzano et al 2014). 30 people with multiple sclerosis were randomised into 2 groups of n=15, undergoing 20 1hr sessions, 3x/week for 9 weeks. The treatment group received active motor

				rehabilitation treatment while the control group received passive mobilisation of the shoulder, elbow, wrist and fingers. Factorial ANOVA with repeated measures (time x group) identified a statistically significant improvement as effect of time was found in both groups for the 9HPT (p=0.0001). (Bonzano et al 2019).26 patients with progressive multiple sclerosis were randomised into active (ATG, n=13) or passive treatment groups (PTG, n=13). Each group underwent 36 1-hr treatment sessions, twice a week. Paired t-tests were used to determine changes between baseline and post-treatment for the 9HPT. A trend towards better performance in the ATG was detected in the 9HPT (p=0.06), however there were no significant differences between baseline and post-treatment in either treatment group. (Boffa et al 2020).
Neurological function assessed with: Purdue Pegboard	0 (1 RCT)[8]	⨁◯◯◯ Very low[a,b,d]	-	37 people with multiple sclerosis were randomised to an intervention (n=19) or control (n=18) group. The experimental group received a home-based upper limb training program, 2 60-min sessions per week for 8 weeks. The control group received information regarding upper limp alterations and a schedule for basic exercises to be performed 2x/week for 60 mins at home for 8 weeks. A two-way analysis of variance tests identified significant between-group difference improvement in the Purdue Pegboard (p<0.01) in the more affected limb (mean difference in intervention group was 2.05, SD 1.60). (Ortiz-Rubio et al 2016).

1. Boffa, G.,Tacchino,A.,Sbragia,E.,Schiavi,S.,Droby,A.,Piaggio,N.,Bommarito,G.,Girardi,G.,Mancardi,G. L.,Brichetto,G.,& Inglese,M. Preserved brain functional plasticity after upper limb task-oriented rehabilitation in progressive multiple sclerosis. European Journal of Neurology; 2020.
2. Grubic Kezele T., Babic M.,Kauzlaric-Zivkovic T.,Gulic T. Combined upper limb and breathing exercise programme for pain management in ambulatory and non-ambulatory multiple sclerosis individuals: part II analyses from feasibility study. Neurological Sciences; 2020.
3. Gervasoni, E.,Cattaneo,D.,Bertoni,R.,Grosso,C.,Bisio,A.,Rovaris,M.,& Bove,M.. Effect of arm cycling and task-oriented exercises on fatigue and upper limb performance in multiple sclerosis: a randomized crossover study. International Journal of Rehabilitation Research; 2019.
4. Grubic Kezele T., Babic M.,Stimac D. Exploring the feasibility of a mild and short 4-week combined upper limb and breathing exercise program as a possible home base program to decrease fatigue and improve quality of life in ambulatory and non-ambulatory multiple sclerosis individuals.. Neurological Sciences; 2019.
5. Bonzano, L.,Pedullà,L.,Tacchino,A.,Brichetto,G.,Battaglia,M. A.,Mancardi,G. L.,& Bove,M. Upper limb motor training based on task-oriented exercises induces functional brain reorganization in patients with multiple sclerosis. Neuroscience; 2019.

6. Bonzano, L.,Tacchino,A.,Brichetto,G.,Roccatagliata,L.,Dessypris,A.,Feraco,P.,Lopes De Carvalho,M. L.,Battaglia,M. A.,Mancardi,G. L.,& Bove,M. Upper limb motor rehabilitation impacts white matter microstructure in multiple sclerosis.. NeuroImage; 2014.
7. Nociti, V. Prosperini L. Ulivelli M. Losavio F. A. Bartalini S. Caggiula M. Cioncoloni D. Caliandro P. Minciotti I. Mirabella M. & Padua L. Effects of rehabilitation treatment of the upper limb in multiple sclerosis patients and predictive value of neurophysiological measures. European Journal of Physical and Rehabilitation Medicine; 2016.
8. Ortiz-Rubio A., Cabrera-Martos I.,Rodriguez-Torres J.,Fajardo-Contreras W.,Diaz-Pelegrina A.,Valenza M.C. Effects of a Home-Based Upper Limb Training Program in Patients With Multiple Sclerosis: A Randomized Controlled Trial.. Archives of physical medicine and rehabilitation; 2016.

a. No participants with FRDA included.
b. Only one study published.
c. Confidence intervals not reported.
d. Small sample size.
e. Allocation not blinded to participants, investigators or treating clinicians.
f. Confidence intervals not reported in 4/5 studies.
g. Participants, treating clinician and investigators not blinded in 4/5 studies.

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)	
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ADDITIONAL CONSIDERATIONS

	Barthel index				60 min/session and performed independent home exercise 3 days/week for 4 weeks, >20mins/session. The no exercise group performed no exercises. A 2-way mixed-model repeated-measures ANOVA (time x intervention) demonstrated no statistically significant interaction in the Barthel Index.
	Quality of life assessed with: Modified fatigue impact scale	0 (3 RCTs)[1,3,4]	⨁◯◯◯ Very low[a,c,e]	-	26 patients with progressive multiple sclerosis were randomised into active (ATG, n=13) or passive treatment groups (PTG, n=13). Each group underwent 36 1-hr treatment sessions, twice a week. The interaction treatment group x time in RM ANCOVA showed a significant improvement, post-treatment versus baseline, in the ATG versus the PTG, in terms of MFIS total (p = 0.03) and physical (p = 0.01) scores. (Boffa et al 2019). 19 individuals with multiple sclerosis were divided into 2 groups: exercise (n=10, controlled group 2 days/week, 60 min/session with independent home exercise 3 days/week, >=20min/session), no exercise (n=9). A two-way mixed-model repeated-measures ANOVA identified a statistically significant group by time interaction on the MFIS scores for the exercise non-ambulatory subjects: physical (p=0.009), psychosocial (p=0.018), total (p=0.0008) scores. (Grubic Kezele et al 2019). 22 people with multiple sclerosis were randomly assigned to Group A (8 week rehabilitation followed by 8 weeks of no intervention), or Group B (same treatment in reverse order). An analysis of covariance showed a statistically significant treatment effect in the MFIS (P = 0.05) without any carryover effect (P = 0.63). MFIS was lower after the treatment (T) compared to the waiting list (WL). The combined differences for Groups A and B between WL–T periods for MFIS was [median and interquartile range (Q1–Q3)] 5.2 (10.7) points. (Gervasoni et al 2019).
	Quality of life assessed with: Visual Analogue Scale for pain	0 (1 RCT)[2]	⨁◯◯◯ Very low[a,b,c,d,e]	-	19 patients with multiple sclerosis were randomly divided into 2 groups: exercise (n=10) and no exercise (n=9). The exercise group exercised with a physiotherapy 2 days/week, 60 min/session and performed independent home exercise 3 days/week for 4 weeks, >20mins/session. The no exercise group performed no exercises. A 2-way mixed-model repeated-measures ANOVA (time x intervention) demonstrated a statistically significant group-by-time interaction in the visual analogue scale for pain only in non-ambulatory (p=0.049) individuals and not in ambulatory individuals (p=0.159).

	Quality of life assessed with: SF-36	0 (1 RCT)[4]	⊕◯◯◯ Very low[a,b,c,d,e]	-	19 individuals with multiple sclerosis were divided into 2 groups: exercise (n=10, controlled group 2 days/week, 60 min/session with independent home exercise 3 days/week, >=20min/session), no exercise (n=9). A two-way mixed-model repeated-measures ANOVA identified a statistically significant group by time interaction on the physical functioning (SF-36) (p=0.014) and general health (SF-36) scores (*p*=0.042) in ambulatory subjects.	
	Neurological function assessed with: 9HPT	0 (5 RCTs)[1,3,5,6,7]	⊕◯◯◯ Very low[a,f,g]	-	22 people with multiple sclerosis were randomly assigned to Group A (8 week rehabilitation followed by 8 weeks of no intervention), or Group B (same treatment in reverse order). An analysis of covariance showed no statistically significant treatment effect in the 9HPT (*p*=0.63) with no carryover effect (*p*=0.67) (Gervasoni et al 2019). 30 people with multiple sclerosis were randomised into 2 groups of n=15, undergoing 20 1hr sessions, 3x/week for 2 months. The treatment group received active motor rehabilitation treatment while the control group received passive mobilisation of the shoulder, elbow, wrist and fingers. Factorial ANOVA with repeated measures (time x group) identified a statistically significant improvement as effect of time was found in time required to complete 9HPT (*p*<0.000001). However, there was no significant time x group interaction found in the 9HPT (*p*=0.98). (Bonzano et al 2014). 30 people with multiple sclerosis were randomised into 2 groups of n=15, undergoing 20 1hr sessions, 3x/week for 9 weeks. The treatment group received active motor rehabilitation treatment while the control group received passive mobilisation of the shoulder, elbow, wrist and fingers. Factorial ANOVA with repeated measures (time x group) identified a statistically significant improvement as effect of time was found in both groups for the 9HPT (*p*=0.0001). (Bonzano et al 2019).26 patients with progressive multiple sclerosis were randomised into active (ATG, n=13) or passive treatment groups (PTG, n=13). Each group underwent 36 1-hr treatment sessions, twice a week. Paired t-tests were used to determine changes between baseline and post-treatment for the 9HPT. A trend towards better performance in the ATG was detected in the 9HPT (*p*=0.06), however there were no significant differences between baseline and post-treatment in either treatment group. (Boffa et al 2020).	
	Neurological function assessed with: Purdue	0 (1 RCT)[8]	⊕◯◯◯ Very low[a,b,d]	-	37 people with multiple sclerosis were randomised to an intervention (n=19) or control (n=18) group. The experimental group received a home-based upper limb training program, 2 60-min sessions per week for 8 weeks	

Pegboard				The control group received information regarding upper limp alterations and a schedule for basic exercises to be performed 2x/week for 60 mins at home for 8 weeks. A two-way analysis of variance tests identified significant between-group difference improvement in the Purdue Pegboard (p<0.01) in the more affected limb (mean difference in intervention group was 2.05, SD 1.60). (Ortiz-Rubio et al 2016).

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a. No participants with FRDA included.
b. Only one study published.
c. Confidence intervals not reported.
d. Small sample size.
e. Allocation not blinded to participants, investigators or treating clinicians.
f. Confidence intervals not reported in 4/5 studies.
g. Participants, treating clinician and investigators not blinded in 4/5 studies.

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	There is very low certainty of evidence as per the evidence profile.	

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)
Activities of daily living assessed with: ABILHAND scale	CRITICAL[a]	⨁◯◯◯ VERY LOW[b,c,d,e,f]
Activities of daily living assessed with: Barthel index	CRITICAL[a]	⨁◯◯◯ VERY LOW[b,c,d,e,f]
Quality of life	CRITICAL[a]	⨁◯◯◯

assessed with: Modified fatigue impact scale		VERY LOW[b,d,f]
Quality of life assessed with: Visual Analogue Scale for pain	CRITICAL[a]	⨁◯◯◯ VERY LOW[b,c,d,e,f]
Quality of life assessed with: SF-36	CRITICAL[a]	⨁◯◯◯ VERY LOW[b,c,d,e,f]
Neurological function assessed with: 9HPT	IMPORTANT[g]	⨁◯◯◯ VERY LOW[b,h,i]
Neurological function assessed with: Purdue Pegboard	IMPORTANT[g]	⨁◯◯◯ VERY LOW[b,c,e]

a. Identified as critical (3/3) and important (3/3) by people with FA and important by expert authors on this topic
b. No participants with FRDA included.
c. Only one study published.
d. Confidence intervals not reported.
e. Small sample size.
f. Allocation not blinded to participants, investigators or treating clinicians.
g. Identified as critical (1/6), important (4/6) and low importance (1/6) by people with FA and important by expert authors on this topic.
h. Confidence intervals not reported in 4/5 studies.
i. Participants, treating clinician and investigators not blinded in 4/5 studies.

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		

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.	The Friedreich's ataxia Clinical Management Guideline Patient and Parent Advisory Panel were asked if the intervention was acceptable (weighing up the balance between benefits, harms and costs). 3/5 indicated intensive (arm and hand) rehabilitation for all people with FA was reasonable; 2/5 indicated the intervention varies or was sometimes reasonable. (Aug 2020)

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

We conditionally recommend intensive upper limb rehabilitation for individuals with Friedreich ataxia in a clinical setting.

Justification

We have conditionally endorsed intensive upper limb rehabilitation for individuals with FRDA based on strong evidence in like populations, the clinical reasoning of experienced clinicians and the potential harm of not providing the intervention.

Subgroup considerations

We consider that intensive upper limb rehabilitation may be particularly beneficial in the early stage of the disease and for individuals with a point mutation.

Research priorities

We strongly recommend conducting studies, ideally randomised controlled trials, of intensive upper limb rehabilitation versus no rehabilitation for individuals with FRDA, to inform clinical practice