The effects & mechanisms of increasing running step rate: a feasibility study in a mixed-sex group of runners with patellofemoral pain

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2 mixed-sex group of runners with patellofemoral pain

3

4   Abstract

5   **Objectives:** To explore feasibility of recruitment and retention of runners with

6   patellofemoral pain (PFP), before delivering a step rate intervention.

7   **Design:** Feasibility study

8   **Setting:** Human performance laboratory

9   **Participants:** A mixed-sex sample of runners with PFP (n=11).

10   **Main Outcome Measures:** Average/worst pain and the Kujala Scale were recorded

11   pre/post intervention, alongside lower limb kinematics and surface

12   electromyography (sEMG), sampled during a 3KM treadmill run.

13   **Results:** Recruitment and retention of a mixed-sex cohort was successful, losing one

14   participant to public healthcare and with kinematic and sEMG data lost from single

15   participants only. Clinically meaningful reductions in average (MD=2.1, d=1.7) and

16   worst pain (MD=3.9, d=2.0) were observed. Reductions in both peak knee flexion

17   (MD=3.7˚, d=0.78) and peak hip internal rotation (MD=5.1˚, d=0.96) were observed,

18   which may provide some mechanistic explanation for the identified effects. An

19   increase in both mean amplitude (d=0.53) and integral (d=0.58) were observed for

20   the Vastus Medialis Obliqus (VMO) muscle only, of questionable clinical relevance.

21   **Conclusions:** Recruitment and retention of a mixed sex PFP cohort to a step rate

22   intervention involving detailed biomechanical measures is feasible. There are

23   indications of both likely efficacy and associated mechanisms. Future studies

24   comparing the efficacy of different running retraining approaches are warranted.

25   **Key Words**

26   Patellofemoral Pain, Running, Biomechanics, Electromyography

27

<u>INTRODUCTION</u>

29   Recreational running positively influences cardiac, (Petrovic-Oggiano, Damjanov,

30   Gurinovic, & Glibetic, 2010) metabolic (Williams, 2014) and mental (Ghorbani, et al.,

31   2014) health. Despite the reported benefits, recreational running is reported to bring

32   about an increased risk of musculoskeletal pain. (Saragiotto, et al., 2014; van Gent,

33   et al., 2007) Overall incidence of musculoskeletal pain amongst recreational runners

34   ranges from 19% to 94%, (van Gent, et al., 2007) with patellofemoral pain (PFP)

35   thought to be the most common. (Taunton, et al., 2002) Specific annual incidence of

36   PFP amongst recreational runners ranges from 4% to 21%, (Noehren, Hamill, &

37   Davis, 2013; Ramskov, Barton, Nielsen, & Rasmussen, 2015; Thijs, Van Tiggelen,

38   Roosen, De Clercq, & Witvrouw, 2007), with overall prevalence in sports medicine

39   facilities suggested to be 17%. (Taunton, et al., 2002)

40

41   Running biomechanics has been reported to be a risk factor for, and associated with,

42   running related PFP. Specifically, peak hip adduction during running has been

43   reported to be significantly higher in female runners who develop subsequent PFP

44   when compared to those who remain asymptomatic. (Neal, Barton, Gallie,

45   O'Halloran, & Morrissey, 2016; Noehren, et al., 2013) In addition, based on our

46   recent meta-analysis, (Neal, et al., 2016) peak hip adduction, peak hip internal

47   rotation and contralateral pelvic drop are also significantly higher in runners with

48   PFP when compared to asymptomatic controls. For neuromuscular function, females

49   with PFP have been reported to have a delayed gluteal onset prior to foot contact

50   and shorter gluteal activation duration compared to asymptomatic controls.

51   (Willson, Kernozek, Arndt, Reznichek, & Scott Straker, 2011)

52

At present, evidence suggests that exercise interventions, whilst effective at reducing symptoms in runners with PFP in the short-term, do not result in full symptom resolution. (Earl & Hoch, 2011; Ferber, Kendall, & Farr, 2011) Moreover, exercise may not derive its effects by way of a kinematic mechanism, as multiple studies have demonstrated that exercise programs designed to increase hip strength do not alter running kinematics thought to be associated with PFP. (Earl & Hoch, 2011; Sheerin, Hume, & Whatman, 2012; Willy & Davis, 2011; Wouters, et al., 2012) This brings into question the ability of an exercise intervention to provide long-term resolution to running related PFP, as it fails to target factors known to be associated with the development and persistence of the condition. It is this premise that originally led to the development of what has been termed running retraining, (Heiderscheit, 2011) or more specifically 'the implementation of any cue or strategy designed to alter an individual's running technique'. (I. Davis, 2005)

66

Reports from observational studies, involving visual and verbal cues to reduce peak hip adduction, indicates running retraining may reduce pain and improve function in female runners with PFP who demonstrate more than 20˚ peak hip adduction during running. (Neal, et al., 2016; Noehren, Scholz, & Davis, 2011; Willy, Scholz, & Davis, 2012) The key limitation of this work is that the results can only be extrapolated to a minority of runners with PFP (i.e. females with high peak hip adduction). In addition, a recently completed randomised controlled trial (RCT) has established efficacy for cues to transition from rearfoot to forefoot strike in combination with a load management running program in a mixed-sex, but again a predominantly female,

76   cohort. (Roper, et al., 2016) The limitation of this study is that cues to transition to a

77   forefoot strike are only applicable to those who rearfoot strike at baseline.

78   Additionally, it is thought that such a change to running mechanics may also be

79   injurious by virtue of the increase in Achilles tendon load that is observed with

80   forefoot strike running compared to rearfoot strike running. (Rice & Patel, 2017) This

81   is reinforced by the fact that 25% (2/8) of the runners in this RCT who transitioned to

82   a forefoot strike pattern reported ankle soreness at follow up. (Roper, et al., 2016)

83

84   It has been reported that cues to increase running step rate do not increase Achilles

85   tendon load (Lyght, Nockerts, Kernozek, & Ragan, 2016) and thus may be a more

86   widely applicable running retraining option to those previously studied. A recent

87   feasibility study has reported that a step rate increase of 10% combined with running

88   in a minimalist shoe was superior to foot orthoses at reducing pain and improving

89   function at 12 week follow up in runners with PFP. (Bonacci, Hall, Saunders, &

90   Vicenzino, 2017) An increase in step rate of 10% has also been reported to

91   favourably alter patellofemoral joint stress in both runners with PFP and

92   asymptomatic runners, (Willson, Sharpee, Meardon, & Kernozek, 2014), though the

93   actual reduction in step length reported was much greater (14%). In addition, no

94   evaluation of symptoms could be reported in this study due to the limitation of the

95   cross-sectional, observational design. Observational work in asymptomatic runners

96   also indicates that more modest increases in running step rate of 5% or 7.5% may

97   still reduce peak hip adduction (Heiderscheit, Chumanov, Michalski, Wille, & Ryan,

98   2011; Willy, et al., 2015), albeit of a smaller magnitudes.

99

100 A recent three-arm RCT (Esculier, et al., 2017) found that a running retraining

101 intervention to increase step rate was no more effective than education focused on

102 load management, or compared to the same education combined with exercise

103 therapy in runners with PFP. Whilst no treatment group had superior outcomes, the

104 step rate intervention did result in significant reductions in both worst and running

105 specific pain. All three groups remained symptomatic at the primary end point (20

106 weeks), and running-related pain was higher (2.5/10) in the step rate group

107 compared to previous studies where hip adduction (0.5/10) (Noehren, et al., 2011;

108 Willy, et al., 2012) and strike pattern (1.0/10) (Roper, et al., 2016) has been targeted.

109 This could be explained by the absence of a faded-feedback protocol to facilitate the

110 retraining intervention, (Irene Davis, 2017) which has been found to be effective by

111 previous studies. (Noehren, et al., 2011; Roper, et al., 2016; Willy, et al., 2012)

112

113 The primary aim of this study was to investigate the feasibility of a pragmatic

114 running retraining intervention, by cueing a 7.5% increase in running step rate using

115 a faded feedback protocol. Specific objectives included (i) the recruitment of an

116 appropriate number of both males and females from a clinical population and (ii) the

117 collection of both symptom and function data to determine an estimate of the

118 effects derived from the intervention. The secondary aim was to investigate the

119 potential kinematic and muscle function mechanisms explaining any effects induced

120 by the intervention.

121

METHODS

123   Participants

124   Ethical approval for this study was granted by the Queen Mary Ethics of Research

125   Committee (QMREC2014/63). All participants provided written informed consent

126   prior to study commencement. Participants were recruited from local sports

127   medicine clinics. Sample size was based on the apriori power analysis conducted by

128   the authors of the previous work on running retraining, (Noehren, et al., 2011; Willy,

129   et al., 2012) leading to a total of 10 participants being sought. Participants were of

130   either sex, currently or previously running a minimum of 10 km/week and aged

131   between 18 and 45 years. To be included, participants were required to have

132   atraumatic retropatellar or peripatellar pain during running and one other activity

133   described by the most recent PFP consensus document, which includes squatting,

134   stair ambulation and jumping. (Crossley, et al., 2016) Patellofemoral symptoms

135   needed to be rated at a minimum of three (out of a maximum of 10) using a

136   numerical rating scale (NRS). Potential participants with patellofemoral instability,

137   previous surgery, tibiofemoral pathology or any pathology (musculoskeletal or

138   otherwise) that precluded running participation were excluded.

139

140   Experimental Protocol

141   Included participants were required to present to the Human Performance

142   Laboratory at Queen Mary University of London. In the presence of bilateral

143   symptoms, the knee that scored highest on the numerical rating scale was analysed.

144   In the presence of equivocal symptoms, the dominant limb that would be used to

145   kick a ball was analysed. (Willy, et al., 2012) Both limbs were not entered into the

146 analysis in the presence of bilateral symptoms given the potential for type I error.

147 (Menz, 2005) Prior to data collection, participants completed the Kujala Scale as a

148 subjective measure of function. (Kujala, et al., 1993) The Kujala Scale is a 13-question

149 appraisal of subjective function in those with PFP, with a score of 100 representing

150 no symptoms and a score of 0 indicating complete disability. Participants were also

151 required to rate their average and worst pain in the past week from 0 to 10 using an

152 NRS. Whilst there is no definitive outcome measure for use with a PFP cohort, the

153 NRS and Kujala Scale are reported to be the most valid and responsive measures for

154 detecting change at time of study commencement. (Crossley, Bennell, Cowan, &

155 Green, 2004)

156

157 <u>Kinematic Measures</u>

158 Participant movement data were collected during running using a four-camera,

159 infrared motion analysis system (CX-1, Codamotion, Charnwood Dynamics Limited,

160 Leicestershire, UK). (Lack, et al., 2014) 24 infrared markers, consisting of eight

161 individual markers and four rigid clusters of four markers, were placed on standard

162 pelvic and lower limb anatomical landmarks using the CAST protocol. (Cappello,

163 Cappozzo, La Palombara, Lucchetti, & Leardini, 1997) Markers from the pelvis frame

164 to the knee joint centre tracked the thigh segment and markers from the knee joint

165 centre to the ankle joint centre tracked the shank segment. Individual markers were

166 applied using double-sided adhesive tape and secured with transparent surgical

167 tape, with the rigid clusters applied using adjustable elastic straps and secured with

168 cohesive self-adherent bandage. Virtual markers were also identified on the femoral

169 epicondyles and the ankle malleoli, to allow for the calculation of relevant joint

170 centers during an upright standing trial. The hip joint centre was estimated as a

171 projection within the pelvis frame using the methods described by Bell et al (Bell,

172 Pedersen, & Brand, 1990) and did not vary between male and female subjects. The

173 knee joint centre was estimated as the mid-point between the femoral epicondyle

174 markers.

175

176 Participants were asked to run in their usual running shoes and self-select their

177 typical 'steady state' running speed on the laboratory treadmill (Kistler Gaitway,

178 Kistler Group, Winterthur, Switzerland). Participants were instructed to run for a

179 total of three kilometers (KM), with the option to cease if symptoms increased to

180 four or greater on the NRS. 10 seconds of data sampled at 200Hz were collected at

181 0.8/1.8/2.8KM, with distance as opposed to time chosen to act as a constant

182 measure across a cohort of participants running at differing speeds. Multiple data

183 collections were completed to increase reliability of gait analysis. (Monaghan,

184 Delahunt, & Caulfield, 2007) Based on between group differences identified in our

185 recent meta-analysis, (Neal, et al., 2016) variables of interest included peak hip

186 adduction, internal rotation and flexion, peak knee flexion and contralateral pelvic

187 drop, given their retrospective association with PFP.

188

189 <u>Electromyography Measures</u>

190 Surface muscle electromyography (sEMG) were collected simultaneously with the

191 kinematic data using a wireless Delsys TRIGNO system (DELSYS Inc., Natick,

192 Massachusetts, USA). Prior to application, participant's skin was marked, shaved and

193 cleaned with an alcohol swab. Self-contained bipolar electrodes were placed at the

194 motor points of the Gluteus Maximus (GMAX), Gluteus Medius (GMED),

195 Semitendinosus (ST) and Vastus Medialis Obliqus (VMO) adhering to SENIAM

196 guidelines. (Hermens, Freriks, Disselhorst-Klug, & Rau, 2000) 10 seconds of sEMG

197 data sampled at 1926Hz were collected at three specific distance points as described

198 above, but were not synchronised to the kinematic data.

199

200 <u>Running Retraining Intervention</u>

201 Participants completed 18 retraining sessions over the course of six weeks. Each

202 week involved a total of three individual runs, equating to 18 runs in total. For the

203 first four weeks, the initial run was completed in a supervised fashion with the

204 primary investigator (BSN). During the retraining sessions, participants were cued via

205 an audio metronome set at 7.5% above their baseline step rate (calculated during

206 data acquisition), based on the previous work of Willy et al (Willy, et al., 2015). The

207 additional two runs each week were completed independently. A faded feedback

208 protocol successfully used previously was adopted. (Noehren, et al., 2011; Willy, et

209 al., 2012) Feedback exposure was gradually reduced and treadmill run time was

210 gradually increased from 10 minutes to 30 minutes (see figure 1), to facilitate skill

211 acquisition. A slower progression from 10-30 minutes was used (18 sessions over six

212 weeks) compared to previous work (8-10 sessions over two to four weeks), to better

213 adhere to contemporary training progression approaches. (Gabbett, 2016) Further,

214 this pace of progression is used clinically in the chosen recruitment centre,

215 minimising ethical issues from varying usual care. For the final two weeks, all

216 completed sessions were performed independently, without any metronome

217   feedback. All data were collected prior to, and after completion of, the running

218   retraining intervention.

Running Retraining Schedule
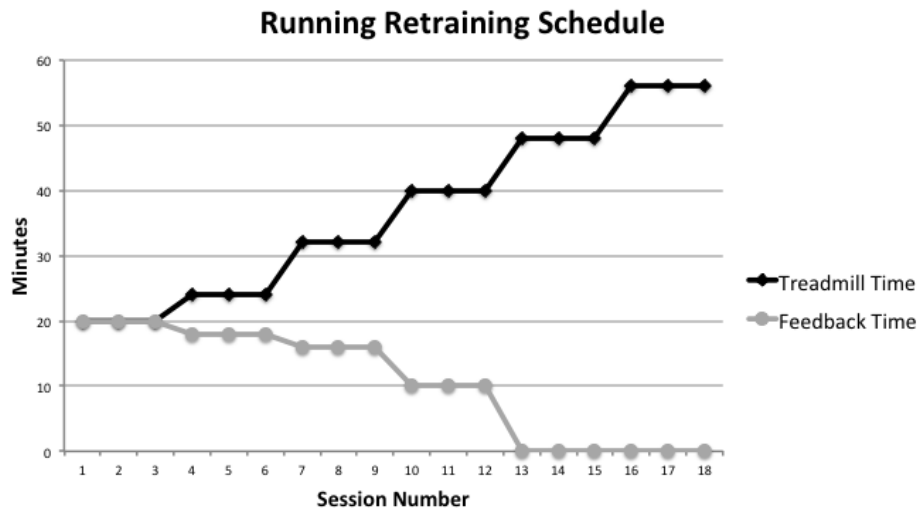
219

220   Figure 1: running retraining schedule depicting the faded feedback protocol
221   employed.
222
223
224   <u>Kinematic Data Analysis</u>

225   Data were analysed offline using a custom written Matlab program (version 2015,

226   Mathworks, Natick, Massachussets, USA). Initial foot contact and toe off were

227   identified using the heel marker on the calcaneal tuberosity and the metatarsal

228   marker on the fifth metatarsal head in the vertical (Z) plane. Consistent with

229   previously described methods, initial foot contact was defined as the point at which

230   the heel marker ceased its descent in the vertical plane. (Zeni, Richards, & Higginson,

231   2008) Toe off was identified using a combination of the heel and metatarsal markers.

232   Specifically, peak acceleration of the metatarsal marker was identified within a

233   specific time point defined by the 70% or greater of the absolute maximum velocity

234   region of the heel marker. (Zeni, et al., 2008) All kinematic data were aligned to

235   initial foot contact, interpolated and normalised to percentage of stride cycle (0% =

236   initial contact, 100% = terminal stance) to facilitate data analysis. Clinical relevance

237   of kinematic data was interpreted with reference to the minimum detectable change

238   data reported by Noehren et al. (Noehren, Manal, & Davis, 2010)

239

240   <u>sEMG Data Analysis</u>

241   sEMG data were processed using an in-built band-pass filter from 25-500 Hz. Raw

242   sEMG data were decomposed using wavelets. (Reaz, Hussain, & Mohd-Yasin, 2006)

243   Post-wavelet decomposition, data were cut into strides using the mean total wavelet

244   power of the VMO muscle, as the typical activation pattern of this muscle

245   (onset/offset) during running is known to align closely to the initial contact (onset)

246   and toe off (offset) phases of running gait. (Flynn & Soutas-Little, 1993) These stride

247   cycle timings were then applied to all sEMG data. Pre and post retraining data were

248   cut into strides independently, but were not used to describe sEMG data as though it

249   were synchronised to the true kinematic gait cycle of the participant. As participants

250   are unlikely to reach signal intensity akin to maximal voluntary isometric contraction

251   (MVIC) during steady state running, data were normalised to the mean of the peak

252   dynamic signal intensity across a single set of strides (0.8KM trial, pre-retraining),

253   which has been reported to be more valid than normalizing to maximal dynamic

254   signal peak. (Bolgla & Uhl, 2007)

255

256   <u>Statistical Analysis</u>

257   All statistical testing were performed offline using Microsoft Excel (Microsoft

258   Corporation, Albuquerque, New Mexico, USA). A Cohen's *d* was calculated to

259   determine the size of all identified interactions, alongside the reporting of mean

260   differences and 95% confidence intervals (CI). Cohen's *d* was interpreted as small ($\leq$

261   0.2), medium (>0.5) and large (>0.8) respectively. (Sullivan & Feinn, 2012) As a

262   feasibility study, not powered apriori to detect statistical significance, dependent

263   sample *t-tests* were not performed and p-values for differences not reported

264   because of the potential for type II error and to avoid giving the impression of there

265   being robust findings from a feasibility study design. The main outcomes were those

266   of recruitment, retention and measurement feasibility.

267

<u>RESULTS</u>

269   A total of 10 (out of 11) participants (four male, six female) completed the study.

270   One female participant was lost to follow up due to a switch of care provision to the

271   National Health Service. Demographics and baseline characteristics of the

272   participants who completed the study are described in table 1.

273

274   <u>Table 1</u>

Variable	Mean (SD)
Sex (Male/Female)	4/6
Age (Years)	31.6 (5.5)
Height (cm)	170.6 (7.8)
Mass (kg)	67.7 (9.8)
Symptom duration (Months)	45.1 (32.1)
Average run volume (KM)	17.0 (9.8)
Step rate (SPM)	163.6 (4.7)
Kujala scale	86.4/100 (6.9)
Average NRS	3.0/10 (1.6)
Worst NRS	6.8/10 (1.5)

275   Participant characteristics

276   *Key: cm=centimeters; kg=kilograms; KM=kilometers; SPM=steps per minute;*

277   *NRS=numerical rating scale.*

278

279

280   <u>Effects</u>

281   Large reductions in both average (d=1.7) and worst (d=2.0) pain were identified post-

282   retraining. The mean difference (MD) of these reductions was 2.1 and 3.9 NRS points

283   respectively and individual participant worst pain responses to the retraining

284   intervention ranged from 1 to 8 NRS points (see figure 2). A modest improvement in

285   function measured with the Kujala Scale was also identified (d=0.12), with a mean

286   difference of 4.4 points.
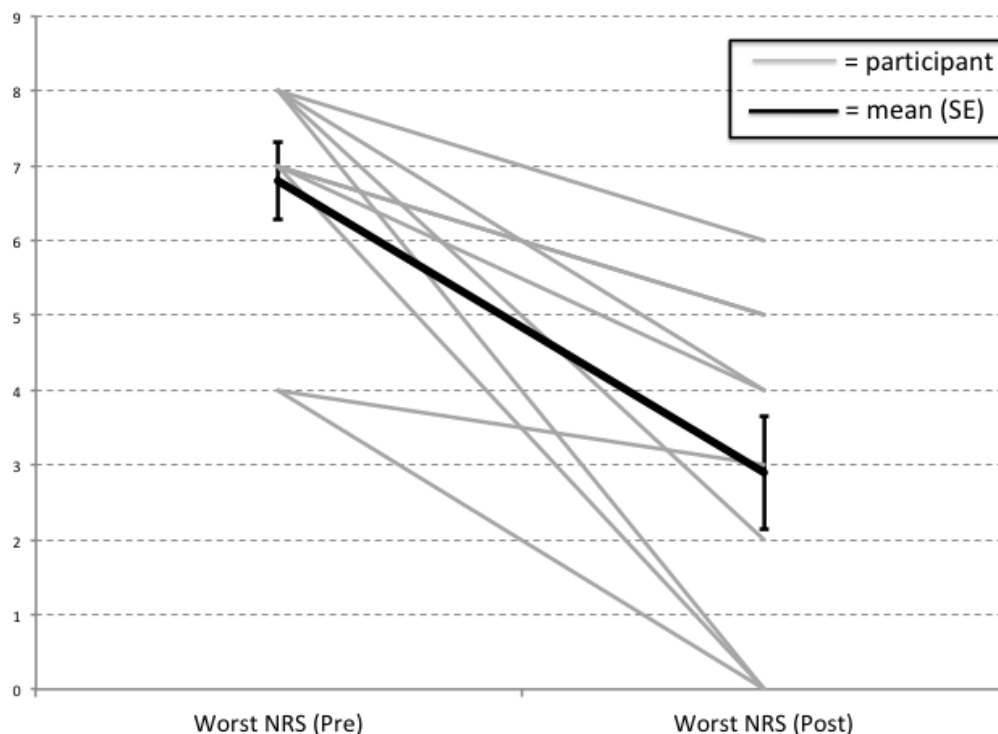
288   Figure 2: mean pooled and individual worst pain responses at baseline (pre) and six
289   weeks follow up (post).

291   <u>Mechanisms</u>

292   *Spatiotemporal*

293   An increase in running step rate at six weeks follow up was observed, with a mean

294   increase of 7.8% (range 2.3% - 11.1%). 3 participants did not achieve a step rate of $\geq$

295   7.5% post retraining.

296

Kinematics

298 One participant was found to have consistently corrupted marker data throughout

299 their trials and was therefore removed from the kinematic analysis. This resulted in a

300 kinematic sample of nine participants (five females, four males). Moderate

301 reductions in both peak knee flexion (MD=3.7˚, *d*=0.78) (see figure 4a) and peak hip

302 adduction (MD=2.4˚, *d*=0.54) (see figure 4b) were identified post-retraining. A large

303 reduction in peak hip internal rotation was also identified post retraining (MD=5.1˚,

304 *d*=0.96) (see figure 4c). A full breakdown of the kinematic analysis can be seen in

305 table 2 and individual participant spatiotemporal and kinematic responses in relation

306 to average/worst pain at six-week follow up are presented in table 3.
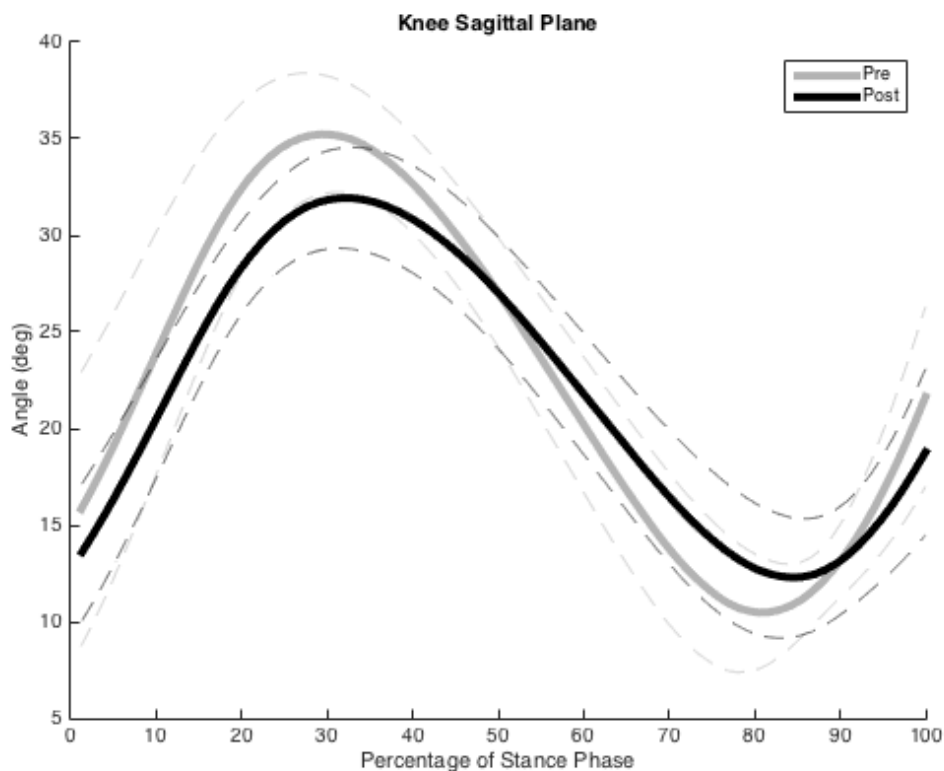
307

308 Figure 4a: mean pattern of hip knee flexion throughout stance at baseline (pre) and
309 six week follow up (post). Knee flexion is positive. Solid line = mean. Dashed line =
310 95% confidence intervals.

Hip Frontal Plane

311
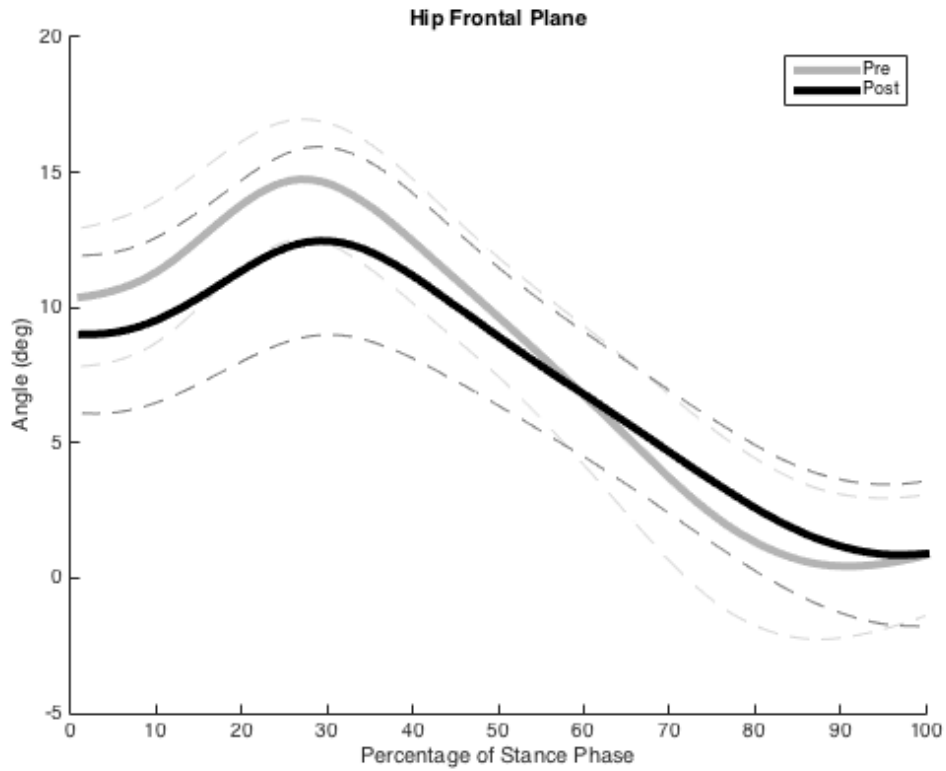
Figure 4b: mean pattern of hip adduction throughout stance at baseline (pre) and six week follow up (post). Hip adduction is positive. Solid line = mean. Dashed line = 95% confidence intervals.

Hip Transverse Plane
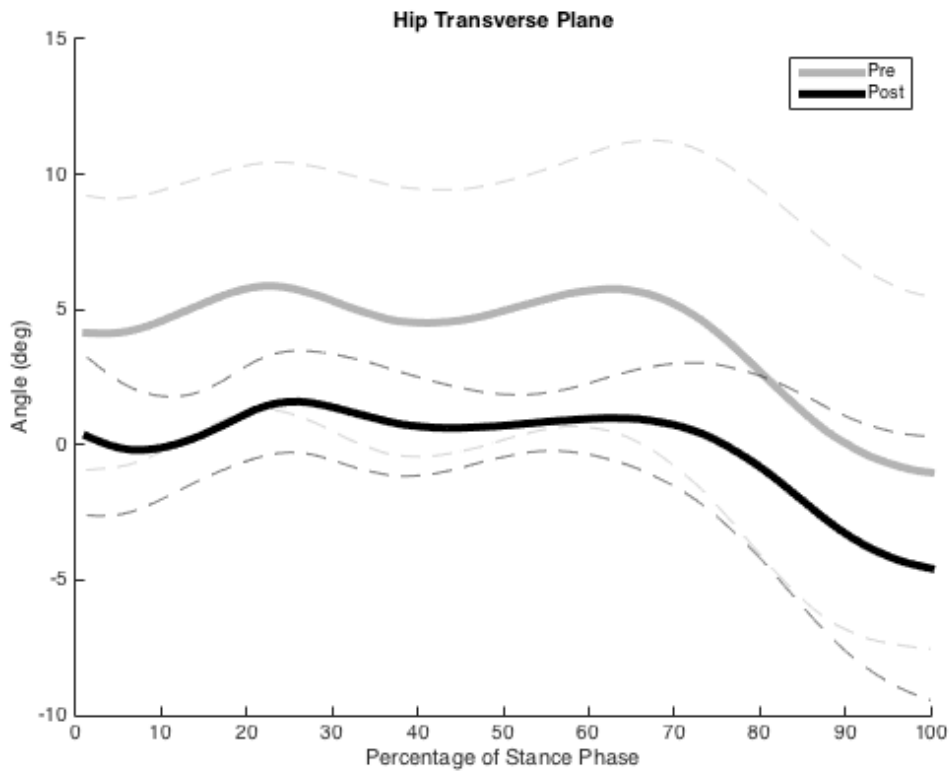
315

316 Figure 4c: mean pattern of hip internal rotation throughout stance at baseline (pre)
317 and six week follow up (post). Hip internal rotation is positive. Solid line = mean.
318 Dashed line = 95% confidence intervals.
319

320 <u>Table 2</u>

Variable	Pre	Post	Mean Difference	95% CI	Cohen's *d*
	Mean (SD)	Mean (SD)			
Average Pain	3.0/10 (1.6)	0.90/10 (0.9)	2.1 (*)	0.88, 3.32	1.7
Worst Pain	6.8/10 (1.5)	2.9/10 (2.3)	3.9 (*)	2.08, 5.72	2.0
Kujala Scale	86.4/100 (6.9)	90.8/100 (5.4)	4.4	-10.22, 1.42	0.1
Peak KFLEX	36.2˚ (5.3)	32.5˚ (4.2)	3.7˚	-1.08, 8.48	0.78
Peak HFLEX	26.7˚ (9.3)	23.1˚ (4.9)	3.6˚	-3.83, 11.03	0.51
Peak HADD	15.6˚ (3.5)	13.2˚ (5.4)	2.4˚	-2.15, 6.95	0.54
Peak CLPD	4.3˚ (2.7)	2.8˚ (2.4)	1.5˚	-1.05, 4.05	0.59
Peak HIR	9.1˚ (7.7)	4.0˚ (2.9)	5.1˚ (*)	-0.71, 10.91	0.96

321 Pre and post retraining means, standard deviations, mean differences, 95%
322 confidence intervals and effect sizes
323

324 *Key: (*)=mean difference exceeds MDC; SD=standard deviation; CI=confidence*
325 *interval; HADD=hip adduction; HIR=hip internal rotation; CLPD=contralateral pelvic*
326 *drop; KFLEX= knee flexion; HFLEX= hip flexion.*
327

Table 3

Participant	Peak KFLEX at Follow Up	Peak HADD at Follow Up	Peak HIR at Follow Up	Peak KFLEX at Follow Up	Baseline Step Rate	Step Rate % Increase	Average Pain at Follow Up (x/10)	Worst Pain at Follow Up (x/10)
1	↓	↓	↓*	↓	160	11.1%	1	6
2	↓*	↓*	↓*	↓*	172	2.3%	2	5
3	↓	↓*	↓*	↓	168	7.7%	0	0
4	↓*	↓*	↓*	↓*	164	8.9%	0	3
5	↓	↑*	↓*	↓	168	7.7%	0	0
6	↓	↓*	↑	↓	164	8.9%	2	2
7	↑	↑	↓*	↑	164	5.7%	0	0
8	↓*	↓*	↑	↓*	158	10.2%	1	4
9	↑	↓	↑*	↑	158	6.0%	2	4

Individual participant kinematic, spatiotemporal and symptom responses to retraining

Key: (*)=difference exceeds MDC; HADD=hip adduction; HIR=hip internal rotation; KFLEX=knee flexion; A-NRS= average pain; W-NRS=worst pain.

328　*sEMG*

329　One participant was found to have consistently corrupted sensor data throughout

330　their trials and was therefore removed from the sEMG analysis. This resulted in a

331　sEMG sample of 9 participants (6 females, 3 males). A mean of peak muscle

332　amplitudes, in addition to an integral (amplitude x duration) of each decomposed

333　signal were calculated for each muscle pre and post retraining. For mean amplitude,

334　minimal changes post-retraining were identified for GMAX (*d*=0.02), GMED (*d*=0.07)

335　and ST (*d*=0.05). However, for VMO, an increase in mean amplitude was observed

336　post-retraining, associated with a medium effect size (*d*=0.53, 95% CI -0.09, 0.03).

337　For muscle integral, a similar interaction was identified, with minimal changes seen

338　post-retraining for GMAX (*d*=0.04), GMED (*d*=0.04) and ST (*d*=0.09). For VMO, an

339　increase was observed, associated with a medium effect size (*d*=0.58, 95% CI -0.06,

340　0.02).

341

DISCUSSION

343 The results of this study suggest that a faded feedback protocol to increase running

344 step rate by 7.5%, is feasible in a clinical setting. A mixed sex cohort was successfully

345 recruited and a low dropout rate (n=1) was achieved. Furthermore, potential

346 clinically relevant changes in both average and worst pain were identified post-

347 retraining, suggesting that the intervention has potential efficacy and warrants

348 further appraisal in an adequately powered RCT.

349

350 The mean reductions in both average and worst pain seen within this study are

351 smaller than those identified by previous running retraining studies, (Noehren, et al.,

352 2011; Roper, et al., 2016; Willy, et al., 2012) although no inference on average or

353 worst pain as individual outcomes were made by these studies and the feedback

354 employed was different. Further, both this feasibility study and the referenced works

355 were essentially underpowered for all but the most preliminary of conclusions.

356 When analysing the reductions in worst pain from this study, only 3/10 participants

357 were asymptomatic at six-week follow up and just one participant had pain $\leq$ 3/10.

358 This means that the 6 remaining participants would continue to be eligible for

359 inclusion into a clinical trial using currently accepted criteria, (Crossley, et al., 2016)

360 meaning that the intervention could be defined as unsuccessful in 60% of our cohort

361 if using worst pain as the primary outcome.

362

363 A recent high quality RCT identified that a 7.5% step rate increase, with the option of

364 transitioning to a forefoot strike pattern if deemed necessary, was no more effective

365 than comparative education or exercise interventions. (Esculier, et al., 2017) When

366   comparing the symptom reductions achieved in this study (6 week follow up) to the

367   relevant time point in the Esculier et al RCT (8 week follow up), (Esculier, et al., 2017)

368   both average and worst VAS are comparable for our step rate intervention compared

369   to all 3 intervention groups (education, exercise plus education, running retraining

370   plus education). It could be suggested that running retraining is in fact a form of load

371   management or graded exposure, which may explain why it was found to be no

372   more effective than education on training loads by Esculier et al. (Esculier, et al.,

373   2017) However, Roper et al (Roper, et al., 2016) reported efficacy of retraining from

374   rearfoot to forefoot strike running. Importantly, this retraining strategy produced

375   larger pain reductions when delivered using a faded feedback protocol, over and

376   above an equivocal progressive duration running protocol. This suggests that a form

377   of feedback is required over and above a load management intervention where

378   there is a clinical need. A further potential explanation for the more modest

379   symptom responses to step rate retraining reported by Esculier et al, (Esculier, et al.,

380   2017) is that feedback is likely to have needed to be subject or subgroup specific

381   and not all participants will have a baseline step rate amenable to an increase.

382

383   Previous studies on running retraining have established a potential kinematic

384   mechanism at the hip to explain their positive effects, specifically a 5˚ reduction in

385   peak hip adduction. (Noehren, et al., 2011; Willy, et al., 2012) The results of this

386   study are in line with this, identifying a smaller but still clinically meaningful mean

387   difference of 2.4˚ that was associated with a moderate effect size (Table 2). Our

388   mixed-sex sample could explain this smaller mean difference, as the previous work

389   of both Noehren et al (Noehren, et al., 2011) and Willy et al (Willy, et al., 2012)

390 purposefully recruited female participants with higher than average peak hip

391 adduction, which may be more amenable to change. However, as our results have

392 identified a reduction in peak hip adduction equivalent to a previous 7.5% step rate

393 increase study in asymptomatic runners, (Willy, et al., 2015) it is suggested that a

394 larger increase in step rate (10%) will result in greater reductions in peak hip

395 adduction equivalent to those seen in asymptomatic runners (Heiderscheit, et al.,

396 2011). A 10% step rate increase is known to reduce both patellofemoral joint stress

397 (Willson, et al., 2014) and pain (Bonacci, et al., 2017) in runners with PFP, whereas a

398 7.5% step rate increase (Esculier, et al., 2017) resulted in non-significant changes in

399 both peak patellofemoral reaction force and average patellofemoral loading rate in a

400 recent RCT. Clinically, it may be sensible to start retraining with a more modest 7.5%

401 step rate increase, increasing to 10% or greater if tolerated, especially in those with

402 low baseline step rate.

403

404 In addition to reducing peak hip adduction, the results of this study have identified

405 two novel potential kinematic mechanisms, being a reduction in both peak hip

406 internal rotation and knee flexion. The identified mean difference in peak hip

407 internal rotation of 5.1˚ is above the MDC of 3.7˚ reported by Noehren et

408 al (Noehren, et al., 2010) and was associated with a large effect size (*d*=0.96). Peak

409 hip internal rotation is associated with running related PFP (Neal, et al., 2016) and

410 can result in increased patellofemoral joint stress by increasing contact pressures at

411 the lateral patellar facet. (Salsich & Perman, 2007) Thus, given the plausibility for

412 reducing hip internal rotation during running gait to favourably alter PFP symptoms

413 and the size of the identified effect, one could argue that a clinically meaningful

414 change has been identified.

415

416 A reduction in peak knee flexion of 3.7˚ is in line with the work of Lenhart et al,

417 (Lenhart, Thelen, Wille, Chumanov, & Heiderscheit, 2014) who reported a reduction

418 in peak knee flexion of 3.3˚ with a 10% step rate increase in a normative cohort.

419 Within this musculoskeletal model, (Lenhart, et al., 2014) peak knee flexion

420 correlates positively with patellofemoral joint force, indicating this finding may be

421 clinically relevant. This effect is likely due to changes in patella contact pressures, as

422 a subsequent modeling study reports that lateral patellar arthrokinematics were not

423 significantly altered by a 10% step rate increase. (Lenhart, et al., 2015) At an

424 individual level, kinematic changes seem to correlate poorly with symptom

425 improvements post-step rate retraining (see table 3). For example, two participants

426 (one male, one female) had an increased peak hip adduction post-retraining (see

427 table 3), with both participants asymptomatic for both average and worst pain

428 variables. For the female participant, the increase in peak hip adduction (6.6˚)

429 exceeds the MDIC (2.6˚) and is thus less likely to be related to measurement error.

430 Future studies should look to investigate alternative potential mechanisms of

431 running retraining, such as kinetic changes, load management or graded exposure.

432

433 Previous observational research investigating increasing step rate by 10% has

434 identified increased quadriceps activation (Chumanov, Wille, Michalski, &

435 Heiderscheit, 2012) comparable to the increase seen within this study. VMO activity

436 is known to be altered in some individuals with PFP (Chester, et al., 2008) and VMO

437 weakness is reported to correlate with lateral patella shift. (Sakai, Luo, Rand, & An,

438 2000) Whilst this study design prohibits inference of causality, this sEMG finding may

439 be associated with the reduction in pain seen post-retraining.

440

441 The lack of change in mean gluteal EMG identified by this study is perhaps not

442 surprising given the work of Willson et al, (Willson, et al., 2011) who report no

443 differences in mean gluteal sEMG when comparing female runners with PFP to

444 matched controls. Willson et al (Willson, et al., 2011) do however report that female

445 runners with PFP demonstrate a shorter GMED activation window and delayed onset

446 prior to foot contact in females with PFP. Additionally, Willy & Davis (Willy & Davis,

447 2013) reported earlier GMED activation and an increased GMED activation duration

448 in a small case series of 2 female runners with PFP post-mirror running retraining.

449 Combined with findings from our study, this indicates that changes to gluteal muscle

450 activation patterns rather than magnitude may provide mechanistic explanation for

451 the reduction in pain. Further research is needed to explore this and a limitation of

452 the current study is the fact that the sEMG were not synchronised to the kinematic

453 system, meaning not all variables of interest from the previous literature could be

454 investigated.

455

456 <u>Future Directions</u>

457 Based on the results of this feasibility study, a future RCT should look to compare a

458 step rate intervention against an exercise therapy control and investigation of effects

459 to long-term follow up (~12 months) is advocated. Future work on running retraining

460 should seek to use a faded feedback protocol, as it appears to result in superior

461 outcomes. Recruitment of participants with a step rate of <160 (>1 SD below the

462 mean of this cohort) who are more likely to be amenable to step rate retraining or

463 stratifying outcome analysis by baseline cadence is worth considering – a strategy

464 that would require greater samples but produce more generalisable findings. Sub-

465 group analysis by baseline kinematic variables associated with PFP such as hip

466 adduction may also be indicated, though kinematic variables do not appear to be

467 sensitive to predicting those who may respond to a step rate intervention.

468

469 Whilst this feasibility trial was not powered apriori to investigate these effects, a

470 post-hoc calculation using the mean difference of both average and worst pain

471 revealed that a sample of 10 participants is adequate to investigate symptom

472 changes post-step rate retraining with adequate statistical power (α=0.05, β=0.20). It

473 is therefore advisable that future trials adhere to the so-called rule of 10, recruiting

474 10 participants per individual variable investigated to minimize risk of bias (Peduzzi,

475 Concato, Kemper, Holford, & Feinstein, 1996) 10% of the biomechanical data in this

476 study was lost due to data corruption and it is advisable that this be factored in to

477 any sample size calculation for mechanistic outcomes in future studies.

478

479 Comparing the results of this study to the previous work on running retraining

480 proved challenging given the heterogeneity of pain outcomes collected. It is

481 advisable that future work collects data on both average/usual and worst/running

482 related symptoms to allow for more clinically meaningful comparisons. The mean

483 difference in the Kujala scale identified falls well below the accepted MCID of 10

484 points (Crossley, et al., 2004) and given the high baseline scores seen in the

485   population studied, a ceiling effect can be suggested. Future studies are advised to

486   consider an alternative measure of subjective function, with the lower extremity

487   functional scale (LEFS), used by previous studies, (Noehren, et al., 2011; Willy, et al.,

488   2012) and the recently developed patellofemoral subscale of the Knee Osteoarthritis

489   Outcome Score (KOOS), (Crossley, Macri, Cowan, Collins, & Roos, 2017) particularly

490   worthy of consideration.

491

492   <u>CONCLUSION</u>

493   The results of this study confirm that increasing running step rate using a faded-

494   feedback protocol is a feasible and effective intervention for use in a mixed sex UK

495   cohort. Future studies should focus on investigating the long-term efficacy of

496   running retraining in a cohort that have a clear treatment target (i.e. low step rate),

497   compared to an appropriate control. A sample size of ten participants per

498   group/variable is adequate to detect minimum clinically important differences with

499   adequate statistical power. In addition to future work establishing efficacy,

500   exploration of both forms of feedback and treatment mechanisms is encouraged.

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502
503
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673

(1) Conflict of Interest

The authors declare that they have no conflicts of interest in relation to this study.

(2) Ethical Approval

Ethical approval was sought and subsequently granted by the Queen Mary Ethics of Research Committee (QMREC2014/63)

(3) Funding