Eur J Vasc Endovasc Surg (2016) 52, 90–98

Diagnosis and Management of Iliac Artery Endofibrosis: Results of a Delphi Consensus Study

INSITE Collaborators (INternational Study group for Identification and Treatment of Endofibrosis) *

Objective: Iliac endofibrosis is a rare condition that may result in a reduction of blood flow to the lower extremity in young, otherwise healthy individuals. The data to inform everyday clinical management are weak and therefore a Delphi consensus methodology was used to explore areas of consensus and disagreement concerning the diagnosis and management of patients with suspected iliac endofibrosis.

Methods: A three-round Delphi questionnaire approach was used among vascular surgeons, sports physicians, sports scientists, radiologists, and clinical vascular scientists with experience of treating this condition to explore diagnosis and clinical management issues for patients with suspected iliac artery endofibrosis. Analysis is based on 18 responses to round 2 and 14 responses to round 3, with agreement reported when 70% of respondents were in agreement.

Results: Initially there was agreement on the typical symptoms at presentation and the need for an exercise test in the diagnosis. Round 3 clarified that duplex ultrasound was a useful tool in the diagnosis of endofibrosis. There was consensus on the most appropriate type of surgery (endarterectomy and vein patch) and that endovascular interventions were inadvisable. The final round helped to inform aspects of the natural history and post-operative surveillance. Progression of the disease was likely with continued exercise but cessation may prevent progression. Surveillance after surgery is generally recommended yearly with at least a clinical assessment.

Conclusions: There is broad agreement about the presenting symptoms and the investigations required to confirm (or exclude) the diagnosis of iliac endofibrosis. There was consensus on the surgical approach to repair. Disagreement existed about the specific diagnostic criteria that should be applied during non-invasive testing and about post-operative care and resumption of exercise.

Article history: Received 18 January 2016, Accepted 6 April 2016, Available online 17 May 2016

Keywords: Cycling, Endofibrosis, External iliac artery, Peripheral artery disease, Sport

INTRODUCTION

The onset of exercise-induced leg pain in young, otherwise healthy individuals can often lead to diagnostic difficulty, particularly when those affected are high-performance athletes. Over recent years it has become apparent that these symptoms could be caused by non-atherosclerotic lesions. Among these are endofibrosis of the arteries specifically of the external iliac artery and, perhaps lengthening and kinking of the external iliac artery.[1,2] There is an increasing awareness of iliac endofibrosis, particularly among elite sports people (especially cyclists) consequent to a number of high profile cases. Yet iliac endofibrosis is a condition that is infrequently managed by vascular specialists and often presents to non-specialists and allied healthcare professionals. Consequently patients with the condition may have a delayed diagnosis or incorrect management.[3]

A recent systematic review of the literature has confirmed there is a paucity of robust data to inform the diagnosis and management of patients with iliac endofibrosis.[4] At present there are no guidelines or standardised pathways of care to ensure otherwise healthy individuals receive consistent advice and appropriate management of their condition.

This study reports the results of a Delphi consensus among members of INSITE (INnternational Study group for Identification and Treatment of Endofibrosis). INSITE is an international inter-disciplinary group of healthcare professionals with a common interest in the management of people with iliac endofibrosis and comprises specialists from sports medicine, sports science, vascular surgery, radiology, and medical imaging. The objective of this study was to identify areas of consensus in diagnosis and management of iliac endofibrosis and to provide a starting point for the future development of guidelines for best practice.

METHODS

The Delphi method is a structured communication technique developed as a systematic, interactive forecasting method. It relies on experts answering questionnaires in two or more rounds. After each round, a facilitator provides

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1078-5884/

http://dx.doi.org/10.1016/j.ejvs.2016.04.004

Table 1. Overall responses to key questions.

		Round 2 ($n = 18$)	Round 3 ($n = 14$)	Frequent comments
1)	**Which of the following points of history are most suggestive of endofibrosis? (mark multiple as desired)**			
a.	Leg swelling	1 (6)	2 (14)	
b.	Leg numbness	3 (17)	4 (31)	
c.	Leg weakness	15 (83)	14 (100)	
d.	Buttock pain	3 (17)	3 (23)	
e.	Thigh pain	14 (78)	14 (100)	
f.	Calf pain	6 (33)	7 (50)	
g.	Involvement of >3 muscle areas	7 (39)	6 (43)	
h.	Resolution of symptoms within 5 min of ceasing exercise	17 (94)	14 (100)	
i.	Absence of back problems	9 (50)	8 (57)	
j.	Other (state):	1 (6)	0 (0)	
2)	**What investigations are required to confirm the diagnosis of iliac endofibrosis?**			
a.	Clinical assessment	12 (67)	10 (71)	
b.	Exercise test	18 (100)	14 (100)	
c.	Duplex ultrasound	12 (67)	10 (71)	
d.	CT angiography	5 (28)	5 (36)	
e.	MRA	4 (22)	4 (29)	
f.	Angiography	3 (17)	3 (21)	
g.	Other (state):	0 (0)	0 (0)	
3)	**Exercise tests are best performed to what level?**			
a.	Until the patient gets symptoms	15 (83)	13 (93)	
b.	Until the patient reaches their anaerobic threshold	3 (17)	1 (7)	
c.	Until the patient reaches predicted max heart rate	1 (6)	0 (0)	
d.	Other (state):	1 (6)	2 (14)	
4)	**How long after stopping exercise should ABPIs (or ankle pressures) ideally be measured?**			
a.	<1 min	11 (61)	13 (93)	
b.	<2 min	1 (6)	5 (36)	
c.	<5 min	0 (0)	0 (0)	
d.	<10 min	4 (22)	1 (7)	
e.	Time-point unimportant	0 (0)	0 (0)	
f.	Other (state):	2 (11)	2 (14)	
5)	**Which of the following indicate a positive exercise test when recorded at the appropriate time after ceasing exercise?**			
a.	Absolute pressure drop			
i.	<10 mmHg	0 (0)[a]	0 (0)	
ii.	10-20 mmHg	4 (25)[a]	3 (21)	
iii.	21-40 mmHg	6 (38)[a]	9 (64)	
iv.	>40 mmHg	2 (13)[a]	5 (36)	
v.	I don't know	2 (31)[a]	3 (21)	
vi.	Other (state):	1 (6)[a]	0 (0)	
b.	**Relative pressure drop between legs**			
i.	<10 mmHg	0 (0)[a]	0 (0)	
ii.	10-20 mmHg	4 (27)[a]	6 (43)	
iii.	21-40 mmHg	6 (40)[a]	11 (79)	
iv.	>40 mmHg	1 (6)[a]	3 (14)	
v.	I don't know	6 (40)[a]	3 (21)	
vi.	Other (state):	1 (6)[a]	0 (0)	
c.	**ABPI**			
i.	<0.4	5 (29)[b]	5 (36)	
ii.	0.4—0.6	8 (47)[b]	7 (50)	
iii.	0.6—0.8	3 (18)[b]	4 (29)	
iv.	>0.8	1 (6)[b]	0 (0)	
v.	I don't know	3 (18)[b]	5 (36)	
vi.	Other (state):	2 (12)[b]	0 (0)	

Continued

Table 1-*continued*

		Round 2 ($n = 18$)	Round 3 ($n = 14$)	Frequent comments
6)	**ABPI should be measured in which of the following positions?**			
a.	On bike with hip flexed	6 (38)[b]	5(36)	
b.	On bike with hip extended	1 (6)[b]	0 (0)	
c.	Supine with hip flexed	1 (6)[b]	1 (7)	
d.	Supine with hip extended	9 (56)[b]	10 (71)	
7)	**What investigations are required in a patient with iliac endofibrosis before operative repair/surgery is performed?**			
a.	Clinical assessment	15 (83)	14 (100)	
b.	Exercise test	17 (94)	14 (100)	
c.	Duplex ultrasound	12 (67)	11 (79)	
d.	CT angiography	6 (33)	6 (43)	
e.	MRA	8 (44)	7 (50)	Cross-sectional imaging may be required especially if non-expert centre or there is diagnostic dilemma post duplex and exercise test
f.	Angiography	4 (22)	4 (29)	
g.	Angiography with provocation (e.g. flexion/ vasodilator)	4 (22)	5 (36)	
h.	Other (state):	0 (0)	0 (0)	
Natural history				
1)	**What contributes most to the development of endofibrosis?**			
a.	Number of hours exercising	14 (78)	11 (79)	Needs further research and it may appear that there is a variability
b.	Intensity of exercise	7 (39)	8 (57)	
c.	Cycling discipline (road/sport/MTB)	8 (44)	7 (50)	
d.	Performance level (e.g. professional/amateur)	6 (33)	4 (29)	
e.	No relationship to volume/intensity/type of exercise	0 (0)	0 (0)	
f.	Age of athlete	1 (6)	1 (7)	
g.	Uncertain	6 (33)	2 (14)	
2)	**Is arterial kinking alone sufficient to cause flow limitation in the absence of endofibrosis?**			
a.	Yes	5 (36)[c]	3 (25)[d]	
b.	No	9 (64)[c]	9 (75)[d]	
3)	**What effect does cessation of exercise have on disease process? (choose one)**			
a.	No effect	0 (0)	0 (0)	
b.	Prevents progression of disease	10 (56)	12 (86)	
c.	Causes regression	0 (0)	0 (0)	
d.	Effect uncertain	9 (50)	9 (64)	
4)	**What effect does continuing exercise have on the disease? (choose one)**			
a.	No effect	0 (0)	0 (0)	
b.	Causes progression at similar rate	11 (61)	11 (79)	Some develop dissection and occlusion — appears rare but important work required to see which ones develop this complication
c.	Hastens progression	3 (17)	3 (21)	
d.	Effect uncertain	6 (33)	9 (64)	
Treatment				
1)	**Which of these are effective in the primary (non-surgical) treatment of endofibrosis?**			
a.	Anti-platelet	0 (0)	14 (7)	
b.	Statin	0 (0)	0 (0)	
c.	NSAID	0 (0)	0 (0)	
d.	No effective medical treatment available	17 (94)	14 (100)	
e.	Other (state):	1(6)	0 (0)	
2)	**Does changing cycling position help?**			
a.	No help	5 (28)	3 (21)	
b.	May improve symptoms	12 (67)	12 (86)	Rarely helps but important to try in the early stages of the disease
c.	May prevent progression of disease	3 (17)	5 (36)	
3)	**Should cyclists be advised to stop cycling as a reasonable first treatment step?**			
a.	Yes	5 (28)	4 (29)	
b.	Yes, but only if non-professional	4 (22)	4 (29)	

Table 1-*continued*

		Round 2 ($n = 18$)	Round 3 ($n = 14$)	Frequent comments
c.	Maybe - assessed on individual patient basis	10 (56)	13 (93)	Difficult to be certain and difficult discussion because good data are not available on the natural history to inform patients
d.	No	1 (6)	1 (7)	
4)	**Who should be offered surgical intervention?**			
a.	Nobody	1 (6)	0 (0)	
b.	Recreational athletes	2 (11)	2 (14)	
c.	Amateur athletes	4 (22)	4 (29)	
d.	Professional athletes	9 (50)	9 (64)	
e.	Anyone with significant impact on QoL	14 (78)	13 (93)	
5)	**Is there a role for endovascular therapy in primary treatment of iliac endofibrosis?**			
a.	Yes - angioplasty only	1 (6)[b]	1 (8)[c]	
b.	Yes - angioplasty and/or stenting	0 (0)[b]	0 (0)[c]	
c.	No	15 (88)[b]	12 (92)[c]	Might serve as a temporary option in athletes
Surgery				
1)	**Shortening alone of the EIA is effective in treating iliac endofibrosis**			
a.	True	0 (0)[c]	0 (0)[e]	
b.	False	14 (100)[c]	11 (100)[e]	
2)	**For patients with endofibrosis apparently limited to EIA on imaging**			
a.	Intervention should be limited to EIA	2 (13)[a]	2 (18)[e]	
b.	Intervention should always extend from EIA to CFA	1 (6)[a]	2 (18)[e]	
c.	Extension is dependent on imaging	7 (47)[a]	7 (64)[e]	
d.	Extension is dependent on intra-operative findings	12 (80)[a]	10 (91)[e]	
3)	**Inguinal ligament release should be considered in most patients undergoing surgery for iliac endofibrosis (may reduce recurrence)**			
a.	True	2 (14)[c]	0 (0)[e]	
b.	False	12 (86)[c]	11 (100)[e]	The inguinal ligament may play a role in the pathogenesis of endofibrosis but difficult to be certain and worries over post-op hernias
4)	**An endarterectomy should always be performed (if technically possible)**			
a.	True	10 (67)[a]	8 (73)[e]	
b.	False	5 (33)[a]	3 (27)[e]	
5)	**Prosthetic patches are preferable to venous patches**			
a.	True	2 (13)[a]	0 (0)[e]	
b.	False	13 (87)[a]	11 (100)[e]	Although vein generally preferable some concerns over late aneurysmal degeneration
6)	**A bypass procedure should be used in most cases of stenosis instead of endarterectomy and patch angioplasty**			
a.	True	0 (0)[a]	1 (9)[e]	
b.	False	12 (80)[a]	10 (91)[e]	
c.	As assessed on individual patient basis	3 (20)[a]	3 (27)[e]	
7)	**Patients with bilateral symptomatic disease are best surgically treated:**			
a.	One side at a time	11 (73)[a]	11 (100)[e]	
b.	Bilaterally at one sitting	0 (0)[a]	0 (0)[e]	
c.	As assessed on individual patient basis	4 (27)[a]	4 (36)[e]	
8)	**Occluded EIA caused by endofibrosis is best treated surgically with:**			
a.	Endarterectomy and patch angioplasty	4 (27)[a]	3 (27)[e]	
b.	Bypass	6 (40)[a]	7 (64)[e]	
c.	Angioplasty	0 (0)[a]	0 (0)[e]	
d.	Angioplasty and stenting	0 (0)[a]	0 (0)[e]	
e.	One of the above based on individual patient assessment	5 (33)[a]	5 (45)[e]	
f.	Other (state):	0 (0)[a]	0 (0)[e]	
Post-operative management				
1)	**The duration of "stand down" (no cycling/sport) post-op should be:**			
	0 weeks	0 (0)[a]	1 (8)[f]	
b.	2-6 weeks	3 (20)[a]	4 (31)[f]	
c.	6-8 weeks	8 (53)[a]	7 (54)[f]	

Continued

Table 1-*continued*

		Round 2 ($n = 18$)	Round 3 ($n = 14$)	Frequent comments
d.	8 weeks-3 months	2 (13)[a]	2 (15)[f]	
e.	3-6 months	1 (6)[a]	1 (8)[f]	
f.	>6 months	0 (0)[a]	0 (0)[f]	
g.	Assessed on an individual patient basis	2 (13)[a]	4 (31)[f]	
2)	**What medication should be given to patients postoperatively?**			
a.	None	1 (6)[a]	0 (0)[d]	
b.	Anti-platelet alone	13 (87)[a]	12 (100)[d]	
c.	Statin alone	1 (6)[a]	0 (0)[d]	
d.	Anti-thrombotic alone (e.g. heparin/coumadin)	0 (0)[a]	0 (0)[d]	
e.	Anti-platelet plus statin	0 (0)[a]	1 (8)[d]	
f.	Statin plus anti-thrombotic	0 (0)[a]	0 (0)[d]	
g.	Anti-thrombotic plus anti-platelet	0 (0)[a]	0 (0)[d]	
h.	Other (state):	0 (0)[a]	0 (0)[d]	
3)	**What is the optimum duration of medication?**			
a.	2 weeks	1 (7)[c]	1 (8)[d]	
b.	2-6 weeks	2 (14)[c]	2 (17)[d]	
c.	6 weeks-3 months	4 (29)[c]	4 (42)[d]	
d.	3 months-1 year	4 (29)[c]	4 (33)[d]	
e.	Life-long	3 (21)[c]	2 (17)[d]	
f.	Other (state):	0 (0)[c]	0 (0)[d]	
4)	**What test is required to establish whether the procedure has been successful?**			
a.	None	2 (11)	2 (15)[f]	
b.	Exercise test	14 (78)	12 (92)[f]	
c.	Non-invasive imaging	9 (50)	5 (38)[f]	
d.	Invasive imaging	0 (0)	1 (8)[f]	
e.	Other (state):	4 (22)	0 (0)[f]	Evaluation of sports results and symptomatology
Long-term follow-up				
1)	**All patients should receive regular surveillance**			
a.	True	14 (78)	12 (86)	
b.	False	4 (22)	2 (14)	
2)	**Surveillance should comprise:**			
a.	Clinical assessment	11 (61)	12 (86)	
b.	Exercise test	7 (39)	6 (43)	
c.	Imaging	12 (67)	9 (64)	Ultrasound can give important information on integrity of patch that would not be available with clinical assessment only
d.	Not applicable (surveillance not required)	2 (11)	3 (21)	
3)	**What is the optimum frequency of surveillance?**			
a.	3 monthly	1 (6)	1 (7)	
b.	6 monthly	2 (11)	1 (7)	
c.	Yearly	11 (61)	11 (79)	
d.	Not applicable (surveillance not required)	2 (11)	2 (14)	
e.	Other (state):	2 (11)	2 (14)	
Management of post-operative complications				
1)	**Re-stenosis should be managed operatively:**			
a.	Only if symptomatic	14 (93)[a]	13 (100)[f]	
b.	If severe stenosis on non-invasive imaging	1 (7)[a]	2 (15)[f]	
c.	Never	0 (0)[a]	0 (0)[f]	
2)	**How is re-stenosis best managed?**			
a.	Angioplasty	5 (36)[c]	5 (45)[e]	
b.	Angioplasty and stenting	2 (14)[c]	2 (18)[e]	
c.	Surgery	7 (50)[c]	10 (91)[e]	
d.	Anti-coagulation	0 (0)[c]	1 (9)[e]	
e.	Cessation of exercise	9 (64)[c]	10 (91)[e]	
f.	Other (state):	2 (14)[c]	1 (9)[e]	
3)	**When should patients have intervention for non-infected aneurysmal dilatation of the EIA?**			
a.	Only if symptomatic	1 (7)[c]	0 (0)[d]	
b.	EIA diameter >1.5 cm	0 (0)[c]	0 (0)[d]	

Table 1-*continued*

		Round 2 ($n = 18$)	Round 3 ($n = 14$)	Frequent comments
c.	EIA diameter >2.0 cm	0 (0)[c]	0 (0)[d]	
d.	EIA diameter >2.5 cm	3 (21)[c]	3 (25)[d]	
e.	EIA diameter >3.0 cm	6 (43)[c]	9 (75)[d]	
f.	Focal aneurysm (pseudoaneurysm)	7 (50)[c]	11 (92)[d]	
g.	Other (state):	5 (36)[c]	2 (17)[d]	
4)	**Once the decision has been made to operate, what is the best method of intervention for patch-related aneurysmal dilatation?**			
a.	Surgery	12 (100)[d]	11 (100)[e]	
b.	Endovascular	0 (0)[d]	0 (0)	
Screening				
1)	**Should routine screening be considered in some populations for the early detection of iliac endofibrosis?**			
a.	Yes	8 (44)	8 (57)	EF may be asymptomatic
b.	No	10 (56)	6 (43)	
2)	**Which population should be screened?**			
a.	None	9 (50)	6 (43)	
b.	Amateur cyclists	2 (11)	1 (7)	
c.	Professional cyclists	9 (50)	8 (57)	
3)	**What is the optimum frequency of screening?**			
a.	Never	9 (50)	7 (50)	
b.	6 monthly	0 (0)	0 (0)	
c.	Yearly	5 (28)	3 (21)	
d.	Every 2 years	2 (11)	4 (29)	
e.	Every 5 years	2 (11)	2 (14)	
4)	**What is the best method of screening?**			
a.	Clinical examination	9 (53)[b]	9 (64)	
b.	Exercise test	14 (82)[b]	14 (100)	
c.	Duplex ultrasound	4 (24)[b]	5 (36)	
d.	CT angiography	0 (0)[b]	0 (0)	
e.	MRA	0 (0)[b]	0 (0)	
f.	Angiography	0 (0)[b]	0 (0)	
5)	**How should patients with asymptomatic endofibrosis be managed?**			
a.	Surgery	0 (0)	0 (0)	
b.	Surgery after discussion	0 (0)	1 (7)	
c.	Surveillance and intervention if evidence of progression	1 (6)	1 (7)	
d.	Surveillance and intervention when symptomatic	10 (56)	14 (100)	
e.	Never intervene	5 (28)	0 (0)	
f.	Advise patient to stop provocative sport	7 (39)	6 (43)	

Data are given as *n* (%).

Underlining refers to consensus (>70%) on a number of issues attained during either the second or third round of the Delphi.

[a] Only 15 people answered this question.

[b] Only 17 people answered this question.

[c] Only 14 people answered this question.

[d] Only 12 people answered this question.

[e] Only 11 people answered this question.

[f] Only 13 people answered this question.

an anonymous summary of the experts' forecasts from the previous round as well as the reasons they provided for their judgements.

The experts are therefore encouraged to revise their earlier answers in light of the replies of other members of the panel. During this process it is anticipated that the range of answers will decrease and the group will converge towards a "correct" answer or "consensus".

A three-round Delphi questionnaire approach was used.[5] The first round of the questionnaire was generated from discussion among experts: vascular surgery ($n = 3$), sports medicine ($n = 2$), radiology ($n = 1$), and a clinical vascular scientist ($n = 1$), and comprised eight broad areas of iliac endofibrosis (with or without iliac kinking) management. Within these eight areas a number of questions with stems were generated (Table 1). Some specialists did not complete specific aspects of the questionnaire that were outside of their area of expertise (e.g. some of the technical aspects of surgery were not necessarily completed by non-surgeons). The second round questionnaire was distributed to the participants who were all members of INSITE in June 2015. They were invited to complete it and provide any

specific comments they perceived to be necessary. The results were collated from the second round. Consensus was achieved when 70% of respondents were in agreement. A third and final round of questionnaires was sent out to the same group of specialists in September 2015. The questionnaire comprised the same questions. Each specialist had access to the second round scores for the whole group as well as their own personal score from the second round, allowing them to compare their views with those of the group and enabling improved evidence for consensus items from the second round. Respondents were invited to re-score the questions in light of the data from the second first round using a Likert scale.[6] The 4-point Likert scale (strongly agree to strongly disagree 1—4) allowed strongly positive responses to be recorded as 1 and strongly negative responses as 4, but has no neutral position and forces a choice. In cases where the respondents' new scores differed from the group score, they were invited to comment to ensure the question was correctly interpreted and expose common fallacies.

RESULTS

In the second round of the Delphi consensus, responses were received from 18 specialists. The overall responses to key questions are shown in Table 1. The third round of the Delphi received responses from 14 specialists and led to consensus on a number of issues (underlined in Table 1), as well as a number of useful comments (also reported in Table 1).

Diagnosis

In the second round there was consensus that leg weakness, thigh pain, and resolution of symptoms within 5 minutes of cessation of exercise were all hallmarks of iliac endofibrosis. After the third round, clinical assessment and duplex ultrasound were thought to be important in the assessment and diagnosis of iliac endofibrosis. The respondents felt that an exercise test (measuring pre- and post-exercise ankle pressure/ABPI) was the most appropriate way to confirm (or exclude) iliac endofibrosis. No specific exercise test is required but there was consensus that the patients should exercise until they reproduced their symptoms. Initially there was no consensus on how rapidly a patient's ankle pressure measurements should be measured after cessation of exercise. However, after round 3, most felt that it was important to measure pressure as quickly as possible (within 1 minute of exercise cessation). There was no consensus on what absolute pressure drop constitutes a positive test for iliac endofibrosis, although a pressure drop between both legs in an individual patient with unilateral symptoms of between 21 mmHg and 40 mmHg was thought to imply a positive test. There was no consensus reached about the absolute level of ABPI at cessation of exercise. There was consensus that measurement of ankle pressures and ABPI was best undertaken with the patient in the supine position. No information was sought on whether manual or automatic blood pressure cuffs were used.

Natural history

In the second Delphi round, there was consensus that the number of hours of cycling was associated with the development of endofibrosis. There was no consensus on whether arterial kinking alone is sufficient to cause flow limitation, or on the effect of continuing or stopping exercise. After the third round, however, there was consensus that arterial kinking in isolation was not responsible for limiting blood flow during exercise and that cessation of exercise prevents the progression of the disease (rather than causing regression) while continuing to exercise leads to disease progression.

Treatment

In the second Delphi round there was immediate consensus that medical therapies are ineffective in the management of endofibrosis, that surgery should be offered to anyone with severely impacted quality of life, and that endovascular interventions have no role in patients with endofibrosis. There was, however, wide variation in views on the most appropriate advice to give to a person with a new diagnosis of iliac endofibrosis.

After the third round there was greater consensus, with agreement that clinicians must decide on an individual patient basis whether cessation of cycling should be considered as a first line of treatment. It was also agreed that changing cycling position may help relieve some symptoms.

Surgery

Shortening of the external iliac artery was thought to be ineffective as a sole strategy for the treatment of endofibrosis. There was consensus that the extent of surgery should be guided by intra-operative findings; however, after the third round of questions pre-operative imaging was thought to be a helpful adjunct in guiding the extent of the procedure. There was consensus that an endarterectomy should be used where possible but that inguinal ligament release, prosthetic patches, and bypass should not be used routinely in surgery. In patients with bilateral disease surgery should be performed one side at a time. The majority of specialists suggested that bypass surgery is generally preferable in patients who present with an occluded external iliac artery, but no consensus was reached.

Post-operative management

There was no consensus on the duration of cessation of exercise/sport ("stand-down") following surgery, although there was consensus that some period of stand-down was required following surgery (54% agreed that a 6—8 week period was most appropriate). Agreement was reached that all patients should receive aspirin following surgery but the duration of treatment remains unclear. There was consensus that surgeons should ideally perform exercise

testing to confirm that the treatment had been successful, but some commented that merely the absence of symptoms was sufficient to suggest that the treatment had been successful.

Long-term follow-up

After the first round there was consensus that all patients should receive regular surveillance. There was no consensus on what form of surveillance patients should receive; however, after the second round of questions there was general consensus that this should at least comprise clinical assessment on a yearly basis. Most (64%) of the respondents suggested that (non-invasive) imaging postoperatively was helpful as it may help to identify pseudoaneurysms or other potential complications that might otherwise be asymptomatic.

Management of post-op complications

In terms of the management of specific local surgical complications there was agreement that a stenosis should only be considered for treatment if symptomatic, that the optimal way to manage these stenoses was surgically (91%), and that patients should be advised to stop exercising (91%). There was a clear indication that intervention for widening or aneurysmal disease subsequent to patch angioplasty was indicated at $>$3.0 cm and for focal pseudoaneurysms, and that these should normally be repaired surgically.

Screening

There was no consensus on the need for screening any groups of asymptomatic individuals for iliac endofibrosis, but 57% suggested that screening in professional cyclists may be justified (an exercise test would be the optimal screening method). If endofibrosis is detected, surgical intervention should be considered only if the patients develop symptoms.

DISCUSSION

There are no guidelines or standard care pathways for the assessment and management of patients with suspected iliac endofibrosis. Iliac endofibrosis is a rare condition and consequently has few published data on which to draw firm conclusions about best practice. Until these data are available it is quite possible that patients may suffer unnecessarily from delayed diagnosis and inappropriate management. In the absence of randomised trials or other controlled studies, the use of Delphi consensus methodology to develop consensus among specialists is a reasonable alternative to inform clinical practice.

There was broad agreement that patients usually present with a symptom complex of leg weakness and thigh pain, typically resolving within 5 minutes of exercise cessation. Non-invasive testing and imaging are usually the first line in the diagnosis, in line with published data.[7] Although an exercise test and duplex ultrasound are helpful in the diagnosis of the condition, the absolute values of pressure drop in the lower limb represent a significant area of controversy with a drop of between 21 mmHg and 40 mmHg being considered diagnostic. However, this controversy may simply reflect that endofibrosis is a progressive disease. In the early stages the symptoms are exclusively at high intensity exercise and the pressure drop is low, in more advanced disease the symptoms commence during lower exercise intensity and are associated with a larger pressure drop. Similarly the duplex criteria for the diagnosis of endofibrosis are poorly defined.[8–10]

A consensus that exercise tests should be standardised was reached, which is an important step forward, and a description of this technique was recently published.[11,12] Patients should be exercised until they develop symptoms and the pressures (ankle and ABPI) should be measured if possible within 1 minute of exercise cessation with the patient in a supine position (and for this some units have adopted automatic systems to measure pressures simultaneously and rapidly in both lower limbs). Some institutions, which consider kinking to be a potential important cause of flow limitation, test the ankle pressures with the patient on a cycle ergometer in an upright position (with a correction being applied for vertical height difference) and with hips flexed.[2]

Management of iliac endofibrosis should always include a careful discussion with the patient and cessation of exercise should always be considered by the athlete before surgery is undertaken. Comments provided by the Delphi participants suggested that these discussions are often difficult as young fit sports people rarely wish to stop exercising. However, this dialogue is important given the paucity of robust surgical outcome data in the short or, more particularly, the long term.[4] Intervention is not without the potential for serious complications and surgery should generally be recommended only for those with symptoms causing a significant impairment of quality of life (and after risks of the procedure have been fully explained).

Original data from the Netherlands had suggested that kinking of the iliac arteries was associated with flow limitation.[2] However, surgical outcome data from the same unit and others suggested that shortening of the iliac arteries in isolation (without endarterectomy and patching) should be performed only in very select cases in which there is no stenotic disease, as, otherwise, this may lead to sub-optimal outcomes.[13] The Delphi consensus corroborated these findings, with most experts suggesting that kinking alone is rarely a cause of significant symptoms and that surgically removing a kink will be largely ineffective if performed as a sole manoeuvre and should be avoided. Although preoperative imaging may provide some guidance on the length of the endofibrotic segment, it was generally agreed that the extent of the endarterectomy should be determined by operative findings. However, comments from some Delphi participants suggested that intra-operative angioscopy also may be useful in guiding the extent of the procedure.

In conclusion, although there was diverse opinion in some areas, consensus was reached on a number of key aspects of iliac endofibrosis. Consensus was reached on the typical presenting symptoms and the non-invasive testing required to confirm the diagnosis. Most experts believe that cessation of exercise prevents the progression of disease and should be recommended to most patients as a first-line management strategy. The type of operation employed should involve an endarterectomy and vein patch repair of the external iliac artery; endovascular therapy and treating kinking alone should be avoided.

The findings of this Delphi consensus study should be used to inform patients with suspected iliac endofibrosis and all healthcare professionals who may be involved in their care. This study has highlighted areas that require further research. As a first step a registry (planned) of patients undergoing surgery will help to capture important data on outcomes that may influence clinical practice. Furthermore, guidelines to standardise care and improve management and outcomes and guide research in the areas of need are warranted.

ACKNOWLEDGEMENTS

The INSITE Collaborators are Professor Robert J Hinchliffe, Bristol, UK, Mr Fabrizio D'Abate, London, UK, Professor Pierre Abraham, Angers, France, Professor Yves Alimi, Marseille, France, Professor Jonathan Beard, Sheffield, UK, Dr Mart Bender, Veldhoven, The Netherlands, Professor Martin Björck, Uppsala, Sweden, Dr Chris Edmundson, Lancaster, UK, Professor Benjamin Fernandez Garcia, University of Oviedo, Spain, Professor Kenneth Cherry, Virginia, USA, Professor Luis Javier Álvarez Fernández, Gijón, Spain, Professor Patrick Feugier, Lyon, France, Professor Jason Lee, Stanford, USA, Dr Roger Palfreeman, Doha Qatar, Professor Louis Passfield, Kent, UK, Mr George Peach, London, UK, Professor Jean-Baptiste Ricco, France, Professor Justin Roake, Christchurch, New Zealand, Professor Hartmut Rimpler, Berlin, Germany, Dr Olivier Rouviere, Lyon, France, Dr Goof Schep, Veldhoven, The Netherlands, Professor Yorck Olaf Schumacher, Doha Qatar, Professor Ian Spark, Adelaide, Australia, and Professor R.E. Zierler, Washington, USA.

CONFLICT OF INTEREST

None.

FUNDING

None.

REFERENCES

1 Chevalier JM, Enon B, Walder J, Barral X, Pillet J, Megret A, et al. Endofibrosis of the external iliac artery in bicycle racers: an unrecognized pathological state. *Ann Vasc Surg* 1986;**1**(3):297—303.
2 Schep G, Bender MH, van de Tempel G, Wijn PF, de Vries WR, Eikelboom BC. Detection and treatment of claudication due to functional iliac obstruction in top endurance athletes: a prospective study. *Lancet* 2002;**359**(9305):466—73.
3 Mansour A, Murney S, Jordan K, LaPerna L. Endofibrosis: an unusual cause of leg pain in an athlete. *J Sports Med Phys Fitness* 2016;**56**(1—2):157—61.
4 Peach G, Schep G, Palfreeman R, Beard JD, Thompson MM, Hinchliffe RJ. Endofibrosis and kinking of the iliac arteries in athletes: a systematic review. *Eur J Vasc Endovasc Surg* 2012;**43**(2):208—17.
5 Murphy MK, Black NA, Lamping DL, McKee CM, Sanderson CF, Askham J, et al. Consensus development methods, and their use in clinical guideline development. *Health Technol Assess* 1998;**2**:1e88.
6 Likert RA. Technique for the measurement of attitudes. *Arch Psychol* 1932;**140**:1—55.
7 Fernández-García B, Alvarez Fernández J, Vega García F, Terrados N, Rodríguez-Alonso M, Alvarez Rodríguez E, et al. Diagnosing external iliac endofibrosis by postexercise ankle to arm index in cyclists. *Med Sci Sports Exerc* 2002;**34**(2):222—7.
8 Alimi Y, Accrocca F, Barthclemy P, Hartung O, Dubuc M, Boufi M. Comparison between duplex scanning and angiographic findings in the evaluation of functional iliac obstruction in top endurance athletes. *Eur J Vasc Endovasc Surg* 2004;**28**(5):513—9.
9 Abraham P, Leftheriotis G, Bourre Y, Chevalier JM, Saumet JL. Echography of external iliac artery endofibrosis in cyclists. *Am J Sports Med* 1993;**21**(6):861—3.
10 Shalhub S, Zierler RE, Smith W, Olmsted K, Clowes AW. Vasospasm as a cause for claudication in athletes with external iliac artery endofibrosis. *J Vasc Surg* 2013;**58**(1):105—11.
11 Abraham P, Bickert S, Vielle B, Chevalier JM, Saumet JL. Pressure measurements at rest and after heavy exercise to detect moderate arterial lesions in athletes. *J Vasc Surg* 2001;**33**(4):721—7.
12 Rouvière O, Feugier P, Gutiérrez JP, Chevalier JM. Arterial endofibrosis in endurance athletes: angiographic features and classification. *Radiology* 2014;**273**(1):294—303.
13 Bender MH, Schep G, Bouts SW, Backx FJ, Moll FL. Endurance athletes with intermittent claudication caused by iliac artery stenosis treated by endarterectomy with vein patch—short- and mid-term results. *Eur J Vasc Endovasc Surg* 2012;**43**(4):472—7.