

Exercise Induced Iliac Artery Endofibrosis in Cyclists

Literature Review

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Epidemiology

Elite athletes, predominantly professional cyclists, can develop arterial flow restriction in one or both legs during exercise. Deformation or progressive stenosis of the iliac artery may reduce blood flow to the lower limb and adversely affect performance (Peach, 2012). The ischemic symptoms are caused by endofibrosis and/or kinking of the external iliac artery (Vink et al., 2008). Iliac artery endofibrosis is a rare cause of arterial stenosis most often found in highly functioning and competitive athletes, commonly cyclists. It is thought to result from repetitive trauma (Weinberg, 2012). Since 1985 reports have been published about claudication at exercise in highly trained cyclists associated with stenosis and or kinking in the iliac artery (Vink et al., 2008). The first two cases of iliac artery endofibrosis were reported in 1985 with prevalence increasing since then with about 20 cases per year since 1990 (Beck, 1995). However, the overall prevalence of iliac flow limitation amongst athletes has been difficult to assess, as the condition may often go unrecognized and data remains sparse (Peach, 2012).

Symptoms

The onset of exercise-induced leg pain in young, otherwise healthy athletes often leads to diagnostic difficulty, especially in high-performance athletes, with most symptoms commonly being attributed to musculoskeletal causes (Peach, 2012). It is often only when symptoms fail to improve regardless of physiotherapy regimens that other potential causes are investigated. Athletes usually complain of an unexplained decrease in performance and thigh discomfort in one or both legs, deep undefined pain, hardening and cramp or an association of these signs (Beck, 1995). Cramp is a predominant symptom with the majority of patients reporting a feeling of swelling, numbness or pain in the calf, thigh or buttock of the affected side (Peach, 2012). At rest, physical examinations may yield normal results but a bruit may be heard over the ipsilateral pelvic fossa or inguinal region in highly trained athletes due to low resting heart rate (Kral et al., 2002). A difference in the intensity or tone of the bruit can be found while peripheral pulses, temperature and colour of the leg remain normal (Abraham et al., 1997). Seldom do patients present with signs and symptoms of restriction of blood supply to tissues in the affected limb (Alimi et al., 2004). This is usually the result of localized dissection and arterial occlusion (Schep et al., 2002)

Regardless of athletes having no cardiovascular risk factors, highly trained young performance athletes were found to have localized flow limitation within the iliac arteries (Peach, 2012). Symptoms are usually unilateral (85%)

with a defined exercise intensity acting as a trigger (Abraham et al., 1997). However, prolonged “sub-trigger” exercise can be performed with recovery facilitated by lowering the exercise intensity to sub-trigger levels (Abraham et al., 1997).

Diagnosis

Delays of 12-41 months in diagnoses have been described between onset of symptoms and correct diagnosis with this delay being greater in professionals than in amateur athletes (Alimi et al., 2004, Schep et al., 2002). The diagnostic difficulty caused by relatively non-specific symptoms has previously led to the development of a detailed questionnaire, which separates vascular from non-vascular causes of leg pain in athletes (Schep et al., 2002). The questionnaire assesses such areas as duration of symptoms, number of muscle groups affected and vascular disease risk factors. Ankle pressure should be determined pre and post exercise with maximal treadmill exercise using diagnostic imaging, along with duplex ultrasonography and contrast angiography, when the leg is flexed at the hip in the cycling position, as this will show concentric stenosis and often lengthening of the affected iliac artery (Alimi et al., 2004). At present, there is no recognized gold standard of imaging against which diagnostic tests can be evaluated (Peach, 2012).

Sensitivity and specificity of physical examinations can be improved with a variety of provocative exercise tests. When resting ankle-brachial pressure index (ABPI) is recorded within five minutes of ceasing maximal exercise it is possible to identify endofibrotic flow-limitation with sensitivity and specificity of up to 100% (Abraham et al., 2001, Bruneau et al., 2009, Fernandez-Garcia et al., 2002, Le Faucheur et al., 2006). In patients with unilateral symptoms, a between leg ABPI difference greater than 0.18 during the first minute of recovery is a significant indicator of an arterial lesion (Taylor & George, 2001). Subtle flow limitations can occur due to kinking rather than endofibrosis and in patients with these limitations, a between leg difference of more than 23mmHg in absolute ankle systolic blood pressure can identify a vascular pathology with a sensitivity of 73% and specificity of 95% (Schep et al., 2002).

Doppler waveforms are often found to be normal other than at extremes of exercise, whilst ultrasound measurement of peak systolic velocity (PSV) may be highly sensitive for endofibrotic stenosis (Ford et al., 2003). Schep et al. found PSV to be significantly higher in affected limbs with the difference accentuated by movements such as hip flexion, isometric psoas contraction and exercise. (Schep et al., 2001, Schep et al., 2002). This allowed identification of arterial kinking and intravascular lesions. Duplex ultrasonography can also be used to

assess anatomical abnormalities of endofibrosis or kinking. In the study by Schep et al., ultrasonography identified external iliac kinking in 39% of symptomatic limbs and intravascular lesions in 61% of symptomatic legs (Schep et al., 2001). After assessing each patient in flexion, extension, with psoas contraction and after exercise, duplex ultrasonography was able to demonstrate abnormalities in the external iliac artery in 82% of symptomatic limbs (Schep et al., 2001). Operative histology has also been used to confirm duplex ultrasound as an identification tool with a sensitivity of 86% (Abraham et al., 1993). The high level of sensitivity is dependent on preselected patient population with relatively severe disease, therefore in studies with patients with less marked symptoms, sensitivity was much lower (von Elm et al., 2007) However, ultrasonography is highly user dependent and may have decreased sensitivity for common iliac artery kinking as this usually occurs in the coronal plane, rendering it difficult to visualise with a conventional ultrasound (Schep et al., 2001).

Static magnetic resonance angiography (MRA) is also useful in assessing iliac flow limitation (Schep et al, 2001, Lim et al., 2009). This must be conducted during hip flexion as even elongated iliac arteries do not tend to kink with hip extension. MRA can identify arterial kinks not visible by ultrasound and allow detailed assessment of iliac artery length, clarifying which patients could benefit from surgical shortening of the artery. Peach (2012) recommends that MRA always be used in conjunction with duplex ultrasound since movement artefact can be substantial.

Static digital subtraction angiography (DSA) has long been considered as the gold standard for assessing flow-limiting atherosclerosis, however, its role in imaging endofibrotic lesions has been less clear. It can be useful for identification of the number and position of any tethering psoas arteries, helping to guide treatment (Alimi et al., 2004). It allows measurement of the pressure gradient across any segment of artery considered to be endofibrotic with the pressure gradient potentially absent at rest but becomes significant following intra-arterial administration of vasodilators (Giannoukas et al, 2006, Maree et al., 2007, Wjiesinghe et al., 2001). CT-angiography is occasionally used with increasing popularity as resolution and 3D reconstruction improves. However, this exposes the patient to radiation and does not offer any significant benefits over MRA combined with Duplex sonography, but it may ultimately be more accessible (Venstermans et al., 2009, Willson et al., 2010).

Aetiology

Most authors suggest that kinking stimulates endofibrosis, which eventually leads to arterial stenosis (Venstermans et al., 2009). Since iliac artery

endofibrosis is most often observed in cyclists, it has been suggested that physical position may contribute to development (Peach, 2012). The aerodynamic position utilized by these athletes leads to hyperflexion at the hip joint and may cause repetitive stretching and deformation of the iliac arteries, which stimulates endofibrotic change (Bender et al., 2004).

Stretching of the artery may be exacerbated by hypertrophy of the psoas muscle and often cyclists with this condition have shown asymmetrical thigh development with thigh circumference being greater on the affected side (Chevalier et al., 1986, Moher et al., 2009). Psoas muscle hypertrophy may increase arterial displacement caused by hyperflexion of the hip thereby heightening kinking and increasing mechanical stresses that may lead to endofibrosis (Bender et al., 2004). Peach et al. (2012) also suggest that repetitive hyperflexion of the hip may cause mechanical trauma to the iliac vessels because of their anatomical fixation to the surrounding tissues. Fascia and collateral branches immobilize iliofemoral vessels as they pass beneath the inguinal ligament and muscular branches to psoas major, preventing the external iliac artery from moving freely during exercise. This makes kinking more acute and increases the traumatic impact of hyperflexion (Peach, 2012). While this kinking may be caused by anatomical fixation, it is also possibly be the result of abnormally long iliac vessels (Schep et al., 2001). Elongation of the artery makes the vessel more tortuous and increases likelihood of kinking during hip flexion (Rousselet et al., 1990). Some investigations suggested that metabolic abnormalities might contribute. Feugier and Chevalier (2004) found nearly 75% of patients who had undergone intervention for symptomatic endofibrosis displayed abnormalities in methionine metabolism. While this may be an incidental association rather than a predisposing factor, further investigation is warranted (Feugier and Chevalier, 2004).

Management

The development of endofibrosis and kinking is multifactorial and hence, management may involve a variety of treatment modalities depending on the relative contribution of individual factors. Conservative measures should be utilised prior to surgical intervention for example, recreational athletes should consider changing sport to one not requiring extreme hip flexion. Long-term outcomes have not yet been established but it has been suggested that untreated stenosis may predispose patients to atherosclerosis later in life (Rousselet et al., 1990). Advice for cyclists should be to reduce time spent on the bike, to raise the handlebars or to bring the saddle position forward to minimize hip flexion and pulling upwards on the pedals should be avoided to reduce psoas hypertrophy (Peach, 2012). While these basic measures may be feasible for recreational

athletes, they often fail to relieve symptoms and may not be realistic for professional cyclists.

Angioplasty has demonstrated some good short-term functional outcomes though patients have all experience recurrence of symptoms within eight weeks, therefore angioplasty is not effective in treating endofibrosis (Giannoukas et al., 2006, Wijesinghe et al., 2001). It also fails to address any underlying abnormalities in vessel length (Feugier & Chevalier, 2004) potentially resulting in dissection of the endofibrotic segment (Cook et al., 1995). Endoluminal stent placement has been found to be an inappropriate treatment as mechanical forces inherent to the condition may cause stent migration or fracture with arterial damage as well as hyperplasia (Schep et al., 2002).

Open surgical correction remains the mainstay of treatment for patients with incapacitating symptoms or those wishing to continue the activity, with surgically removed specimens revealing intimal wall thickening made up of loose connective tissue, the phenomenon of Endofibrosis (Rousselet et al., 1990). Schep et al. (2002) demonstrated that release of restrictive fibrous tissue and division of tethering branches of the external iliac artery can benefit patients with abnormal vessel fixation and arterial kinking. An advantage of this is that the procedure can be performed through a small supra-inguinal incision with the iliac vessels left intact meaning subsequent complications are less likely (Schep et al., 2002). Careful selection of patients is crucial to ensure arterial release is not attempted in isolation for patients who also have significant endoluminal narrowing (Alimi et al., 2004). If diagnostic imaging demonstrates significant arterial elongation, this procedure may not suffice as hip flexion is likely to result in artery kinking, even if the vessel is not abnormally tethered (Schep et al., 2002). Where fibrotic stenosis occurs but arterial length is within the normal range, a conventional endarterectomy may be performed with most groups then achieving closure of the arteriotomy using vein patch angioplasty (Kral et al., 2006, Korsten-Reck et al., 2007, O'Ceallaigh et al., 2002, Wille et al., 1998).

Some authors favour complete resection of the stenosed segment and replacement with a saphenous vein interposition graft (Alimi et al., 2004) while others use PTFE or Gore-Tex grafts when the saphenous vein was too small to be utilized, however no follow up data was presented for these interposition techniques (Abraham et al., 1997, Scavée et al., 2003, Venstermans et al., 2009, Wilson et al., 2010).

Though overall complication rate is perhaps low, the nature of these procedures means that if complications arise they may be catastrophic. Therefore, surgical intervention, particularly interventions involving

reconstruction of the iliac vessels, should only be taken after patients have tried conservative measures and carefully considered all other options, such as changing sport (Peach, 2012).

Discussion

Very little evidence exists of trials with large groups; most evidence derives from isolated case reports and small case series. Arterial kinking has been heavily linked to the genesis of endofibrosis, although Schep et al. suggest iliac kinking alone causes flow limitation. They also found that 48% of patients experience only moderate improvements following surgical release of tethered iliac arteries, suggesting the effect of kinking and vessel stenosis ultimately lead to flow limitation and symptom development. The exact relationship between kinking and endofibrosis remains unclear.

From the studies detailed above, it is clear that although there is potentially useful imaging techniques and treatment samples, neither clinical evaluation or any single form of imaging is sufficient to identify iliac endofibrosis. Also, while endofibrosis can be proven at operation, kinking related flow restrictions are often very difficult to verify since they are often functional rather than anatomical abnormalities (Almi et al., 2004, Bruneau et al., 2009, Feugier & Chevalier, 2004, Le Faucheur et al., 2006).

By considering iliac artery endofibrosis as a potential diagnosis when patients present with these symptoms, physicians can use a discriminatory and physical examination to identify nearly 80% of those with flow limitation (Schep et al., 2002). These patients can then be referred to a physician for duplex imaging, and more detailed imaging to investigate if a flow limitation is present to characterize the lesion. Surgery should only be considered when patients show arterial kinking or intravascular lesion and conservative measures have been considered and disregarded. Information on long-term surgical outcomes is extremely limited, as no comparative studies have been conducted. Although this condition has been gaining increasing recognition, it still often goes undiagnosed or is ineffectively treated as diagnostic techniques for flow limitation lack sensitivity and the lack of consistent methodology and follow up data has led to confusion. Specifically designed tests need to be included in order to diagnose these patients and to ensure effective treatment and management of iliac artery endofibrosis.

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August 25, 2014

References:

- Abraham P, Chevalier JM, Saumet IL. External iliac artery endofibrosis: a 40-year course. *J Sports Med Phys Fitness*. 1997; 37: 297-300.
- Abraham P, Bickert S, Vielle b, Chevalier J, Saumet J. Pressure measurements at rest and after heavy exercise to detect moderate arterial lesions in athletes. *J Vasc Surg*. 2001; 33: 721-727.
- Abraham P, Leftheriotis G, Bourre Y, Chevalier IM, Saumet JL. Echography of external iliac artery endofibrosis in cyclists. *Am J Sports Med*. 1993; 21: 861-863.
- Abraham P, Saumet JL, Chevalier JM. External Iliac Artery Endofibrosis in Athletes. *Sports Medicine*. 1997; 24: 221-226.
- Alimi YS, Accrocca F, Barthelemy 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 VascEndovasc Surg*. 2004; 28: 513-519.
- Beck F and Chevalier JM. Blood Endofibrosis: Typical and Atypical forms: About 164 Cases. *Universite Claude Bernard*, 1995.
- Bender MHM, Schep G, de Vris WR, Hoogeveen AR, Wijn PF. Sports-related flow limitations in the iliac arteries in endurance athletes: aetiology, diagnosis, treatment and further developments. *Sports Med*. 2004; 34: 427-442.
- Bruneau A, Le Faucher A, Mahe G, Vielle B, Leftheriotis G, Abraham P. Endofibrosis in athletes: is a simple bedside exercise helpful or sufficient for the diagnosis? *Clin J Sport Med*. 2009; 19: 282-286.
- Chevalier JM, Enon B, Walder J, Barral X, Pillet J, Megret A et al. Endofibrosis of the external iliac artery in bicycle racers: an unrecognised pathological state. *Ann Vasc Surg*. 1986; 1: 297-303
- Cook P, Erodes L, Selzer P, Ribera F, Palmaz J. Dissection of the external iliac artery in highly trained athletes. *J Vasc Surg*. 1995; 22: 173-177.
- Feugier P, Chevalier J. Endofibrosis of the iliac arteries: an underestimated problem. *ActaChir Belg*. 2004; 104: 635-640.
- Fernandez-Garcia B, Alvarez-Fernandez J, Vega G. Diagnosing external iliac endofibrosis by postexercise ankle to arm index in cyclists. *Med Sci Sports Exerc*. 2002; 34: 222.
- Ford S, Reham A, Bradbury A. External iliac endofibrosis in endurance athletes: a novel case in an endurance runner and a review of the literature. *Eur K VascEndovasc Surg*. 2003; 26: 629-634.
- Giannoukas A, Berczi V, Anoop U, Cleveland T, Bearcd J, Gaines P. Endofibrosis of iliac arteries in high-performance athletes: diagnostic approach and minimally invasive endovascular treatment. *Cardiovasc Inter Rad*. 2006; 29: 866-869.
- Korsten-Reck U, Rocker K, Schmidt-Truckass A, Schumacher YO, Striegel H, Rimpler H et al. External iliac artery occlusion in a young female cyclist. *J Sports Med Phys Fitness*. 2007; 47: 91-95.
- Kral CA, Han DC, Edwards WD, Spittell PC, Tazelaar HD, Cherry KJ Jr. Obstructive external iliac arteriopathy in avid bicyclists: new and variable histopathologic features in four women. *J Vasc Surg*. 2002; 36: 565-570.
- Le Faucheur A, Noury-Desvaux B, Jaquinandi V, Louis Saumet J, Abraham P. Simultaneous arterial pressure recordings improve the detection of endofibrosis. *Med Sci Sports Exerc*. 2006; 38: 1889-1894.
- Lim CS, Gohel MS, Shepherd AC, Davies AH. Iliac artery compression in cyclists: mechanisms diagnosis and treatment. *Eur J VascEndovasc Surg*. 2009; 39: 180-186.
- Maree AO, Ashequal Islam M, Snuderl M, Lamurgalia GM, Stone JR, Olmstead K et al. External iliac artery endofibrosis in an amateur runner: hemodynamic angiographic, histopathological evaluation and percutaneous revascularization. *Vasc Med*. 2007; 12: 203-206.
- Maree AO, Ashequul Islam M, Snuderl M, Lamurgalia GM, Stone JR, Olmsted K, Rosenfield KA, Jaff MR. External iliac artery Endofibrosis in an amateur runner; hemodynamic, angiographic, histopathological evaluation and percutaneous revascularization. *Vasc Med*. 2007; 12:203-206.

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Moher D, Liberati A, Tetzlaff J, Altman DG, Group TP. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Open Med.* 2009; 123-130.

O'Ceallaigh P, Burns P, McLaughlin R, Leader M, Bouchier-Hayes D. Complete external iliac artery occlusion in a 34-year old cyclist. *Eur J VascEndovasc Surg.* 2002; 23: 376-377.

Peach G, Schep G, Palfreeman R, Beard JD, Thompson MM, Hinchliffe RJ. Endofibrosis and Kinking of the Iliac Arteries in Athletes: A Systemic Review. *European Journal of Vascular and Endovascular Surgery.* 2012; 43: 208-217.

Rousselet JP, Saint-Andre P, L'Hoste P, Enon B, Megret A, Chevalier JM. Stenotic intimal thickening of the external iliac artery in competition cyclists. *Hum Pathol.* 1990; 21: 524-529.

Scavée V, Stainier L, Deltombe T, Theys S, Delos M, Trigaux JP et al. External iliac artery endofibrosis: a new possible predisposing factor. *J Vasc Surg.* 2003; 38: 180-182.

Schep G, Bender M, van Tempel G, Wijn P, de Vries W, Eikelboom B. Detection and treatment of claudication due to functional iliac obstruction in top endurance athletes: a prospective study. *Lancet.* 2002; 359: 466-473.

Schep G, Kaandorp DW, Bender MH, Weerdenburg H, van Engeland S, Wijn PF. Magnetic resonance angiography used to detect kinking in the iliac arteries in endurance athletes with claudication. *Physiol Meas.* 2001; 22: 475-487.

Schep G, Schmikli SL, Bender MH, Mosterd WL, Hammacher ER, Wijn PF. Recognising vascular causes of leg complaints in endurance athletes. Part 1: Validation of a decision algorithm. *Int J Sports Med.* 2002; 23: 313-321.

Schep G, Schmikli SL, Bender MH, Mosterd WL, Hammacher ER, Wijn PF. Recognising vascular causes of leg complaints in endurance athletes. Part 2: the value of patient history, physical examination, cycling exercise test and echo-Doppler examination. *Int J Sports Med.* 2002; 23: 322-328.

Schep G, Bender MH, Schmikli SL, Wijn PF. Color Doppler used to detect kinking and intravascular lesions in the iliac arteries in endurance athletes with claudication. *Eur J Ultrasound.* 2001; 14: 129-140.

Taylor A, George K. Ankle to brachial pressure index in normal subjects and trained cyclists with exercise-induced leg pain. *Med Sci Sports Exerc.* 2001; 33: 1862.

Venstermans C, Gielen JL, Salgado R, Bouquillon P, Lauwers J. Endofibrosis of the external iliac artery. *Belgian Journal of Radiography.* 2009; 92: 184-5.

Vink A, Bender MH, Schep G, van Wichen DF, de Weger RA, Pasterkamp G, Moll FL. Histopathological comparison between Endofibrosis of the high-performance cyclist and atherosclerosis in the external iliac artery. *Journal of Vascular Surgery.* 2008; 48: 1485-1463.

Von Elm E, Altman DG, Egger M, Pocock SJ, Gøtzsche PC, Vandenbroucke JP et al. The Strengthening the Reporting of Observational Studies in Epidemiology statement: guidelines for reporting observational studies. *Lancet.* 2007: 1453-1457.

Weinberg I, Jaff M. R, 2012. Nonatherosclerotic Arterial Disorders of the Lower Extremities, *Circulation*, 126, 213-222.

Wilson TD, Revesz E, Podbielski FJ, Blecha MJ. External iliac artery dissection secondary to endofibrosis in a cyclist. *J Vasc Surg.* 2010; 52: 219-221.

Wijesinghe LD, Coughlin PA, Robertson I, Kessel D, Kester RC. Cyclist's iliac syndrome: temporary relief by balloon angioplasty. *Br J Sports Medicine.* 2001: 70-71.

Wille J, de Jong J, Moll F. Endofibrosis of the external iliac artery in sportsmen clinical review and four new cases. *VascEndovasc Surg.* 1998; 32: 323-328.

Wilson TD, Revesz E, Podbielski FJ, Blecha MJ. External iliac artery dissection secondary to endofibrosis in a cyclist. *J Vasc Surg.* 2010; 52: 219-221.