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External Iliac Artery Endofibrosis: A Systematic Literature Review and a Report of Two Cases

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Abstract

Background: External iliac artery endofibrosis is a non-atherosclerotic disease. It affects young individuals, particularly cyclists.

Materials and Methods: A systematic literature review was conducted to highlight the intriguing aspects of this pathology and a total of 493 patients were retrieved to whom we added two patients treated at our institution.

Results: 434 (87%) of the patients were cyclists, 7 (3.6%) triathletes (practicing cycling, running, and swimming), (1.4%) runners, and 18 (8.2%) other. Both limbs were equally involved, and no dominant limb was noted. Twenty-four works and fifty-eight patients were analyzed; in 51.7% (n=30) of cases, the left lower limb was involved, and in 48.3% (n=28) of cases, the right lower limb was involved. The external iliac artery was involved in 90% of cases, however, 30 patients (10%) presented an unusual location, isolated or associated with the lesion beginning on the common iliac artery (5%) or extending to the femoral artery. In addition, the quadricipital artery and the profunda femoris artery can be affected, modifying the haemodynamic conclusions of the complementary examinations Professional cyclists who annually cover an average distance of 10,400 kilometres (range 4,200 min. - 27,500 max. kilometres) were preferentially affected. While the total mileage accumulated since the start of training is a factor that should be considered, we believe that the intensity of the training plays an even more crucial role in the development of the lesion. Noticeably, only 10 (2%) patients had specific atherosclerotic risk factors such as cigarette smoking or mild dyslipidemia. The main symptoms were buttock claudication and calf pain (37% and 30%, respectively). Historically angiography was the diagnostic method of choice in 53% of cases; however, in the recent decades CT and MRI were preferentially used. 71% of the patients were treated with endofibrosectomy, a patch was added in 13 (3%). An Iliofemoral bypass graft was performed in 29% of the patients. Five-year primary and secondary patency rates either for endofibrosectomy or bypass graft reached 90%.

Conclusions: Surgical treatment is the gold standard. Patients unwilling to undergo surgery may opt for less invasive treatments such as percutaneous transluminal angioplasty with stent positioning which seems however, to have suboptimal long-term results.

Keywords: Endofibrosis; External iliac artery, Vascular disease, Athlete, Cyclist

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Introduction

Arterial endofibrosis first described in the mid '80 is a diagnostic challenge for surgeons and clinicians. It is a disease with an insidious clinical presentation that requires some knowledge specifically anamnestic, to be suspected. Arterial endofibrosis is a non-atheromatous vascular pathology preferentially involving young athletes, mostly cyclists. Patients show no clinical history of cardiovascular diseases or risk factors. The elective site of lesions is the external iliac artery in 90% of cases [1,2], and, at pathology, an intimal subendothelial paucicellular fibrosis leading to wall thickening and reduction of the artery lumen caliber up-to the occlusion is shown. This pathologic process explains symptomatology, which in most cases is claudication and pain after maximal exercise as for lower limb chronic atherosclerotic ischemia. We performed a systematic review of the international literature adding our experience on two cases, focused on the demographic aspects, the diagnostic methods, and the therapeutic options.

Case Presentation

Case 1

A 56-year-old Caucasian male patient presented at our Institution for right calf pain after intense physical training. The patient denied cardiovascular risk factors such as hypertension, cigarette smoking, dyslipidaemia or diabetes. He was a former professional cyclist for 20 years but currently he continued to practice intense sport activities, i.e. running (40 km/week) and cycling (120 km/week). His Body Mass Index (BMI) was 23. Limb physical examination was normal, temperature and color were within the normal range, but femoral and popliteal right pulses were hypo-sphygmic. ABI index was 0.8 at rest. A CT scan demonstrated the occlusion of the right external iliac artery extending up-to the origin of epigastric artery (Figure 1). The patient refused an open surgical treatment and therefore an endovascular procedure was performed after patient' risk acceptance. Before the procedure an arteriography of the right ileo-femoral axis was performed. There was an obstruction from the origin of the right external iliac artery to the origin of the right common femoral artery. The artery was then recanalized and a balloon expandable metal stent was placed. The device is a vascular metal stent VIABAHN 8 × 50 and 8 × 100 mm The postoperative course was uneventful, and the patient was placed under double anti-aggregating therapy. At one year follow-up, the CT angiography shows the correct positioning of the stent, with no pathological stenosis observed at this level, the patient is in good health, denies symptoms and performs regular follow-up at our Institution but he significantly reduced his sport activities.

Case 2

A 42-year-old Black (North African) male patient presented at our Institution for left buttock pain after moderate physical training (30 km/week). The patient had no cardiovascular risk factors or associated diseases. He was a retired sprinter (100 mt). BMI was 24. Limb physical examination was normal, temperature and color were within the normal range, but femoral and popliteal right pulses were absent. ABI index was 0.7 at rest. A CT scan demonstrated the occlusion of the left common and external iliac artery. The common, internal, and external iliac arteries were prepared through an extraperitoneal approach. A left common iliac common femoral artery interposition graft (Dacron 8 mm in diameter) was performed. Our follow-up consisted of an echocolorDoppler performed every 6-month and a CT-scan performed yearly. At 3-year follow-up no restenosis and a patent graft were observed. The postoperative course was uneventful. Five years after surgery, the patient is alive and denies specific lower limb symptoms. He denies physical activity and his BMI increased significantly reaching 38.

Methods

The literature search was performed 1 March 2024 on the following databases: PubMed (www.ncbi.nlm.nih.gov/pubmed), SCOPUS (www.scopus.com) and Web of Science (WOS) (www.webofscience.com) to identify all eligible studies. The combination of the following words was used: "endofibrosis", "external iliac artery", and "lower limb". Publications were selected according to the most frequently western countries spoken languages (English, Spanish, Portuguese, Italian, French and German). The selected studies' title and abstracts were independently screened by two Authors (D.S. and P.S.); successively full-text articles of potentially relevant studies were evaluated independently by the same two Authors. When overlapping was found between articles published by the same authors or centers, only the most recent paper was enclosed to avoid duplication. The PubMed function "related articles" and Google Scholar database were used to find further articles. A search on Google book was performed for the analysis of the grey literature (https://books.google.com accessed on 1 March 2024). Studies were included if they contained adequate information regarding patients' demographics, symptoms at presentation, arterial involvement, the type of treatment and follow-up. In the case of duplicate publications, the latest and most complete study was selected.

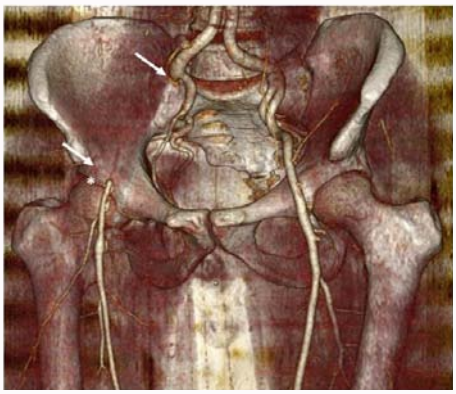

Figure 1: Complete occlusion of the right external iliac artery (arrows) from its origin up-to the epigastric artery origin (asterisk).

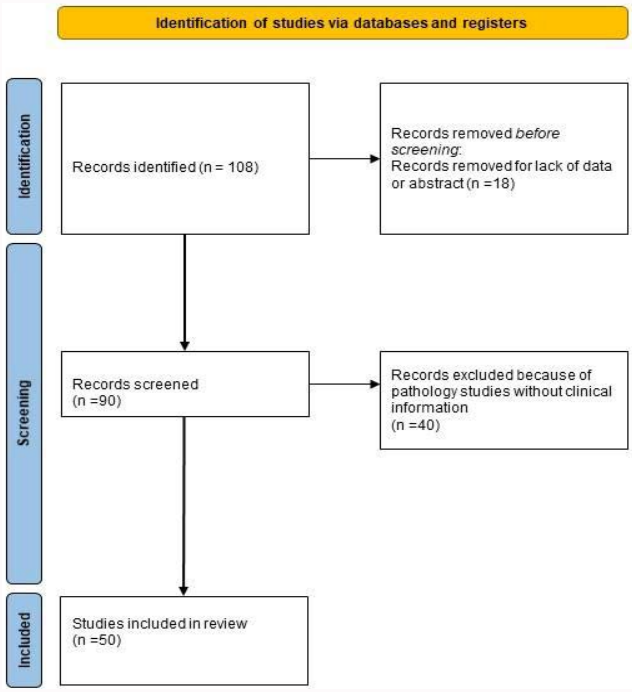

Figure 2: PRISMA Flow Chart.

The PRISMA flow chart [3] for systematic review is schematically reported in Figure 2. Briefly, after the screening for relevance, 50 [1,2,4-15] articles which included 25 [13-50] case reports, 23 [1,4-12,14-30] small series and 2 [2,51] systematic literature reviews form the basis of the present analysis. Most of the studies were performed in USA (18% to 36%) or Europe (29% to 58%), followed by North Africa (1% to 2%), India (1% to 2%) and Canada (1% to 2%).

Results

We were able to retrieve a total of 493 patients to whom we added our personal experience on 2 patients making a total of 495 patients. 421 patients were males, and 74 females. In our analysis we provided an actual estimate of epidemiology of endofibrosis of the external iliac artery which is a non-atherosclerotic vascular pathology causing thickening of the vessel wall and subclinical blood flow obstruction. it affects young athletes, with an average age of 35.6 ± 9.96 years (range 18 min. – 59 max. years, median 39 years, IQR 14.3). We observed that 434 (87%) of the patients were cyclists, 7 (3.6%) triathletes (practicing both cycling and running), (1.4%) runners, and 18 (8.2%) practicing other sport activities. Both limbs were equally involved, and no dominant limb preference was noted. This condition clearly seems to affect predominantly professional cyclists who annually cover an average distance of 10,400 kilometres (range 4,200 min. -27,500 max. kilometres). In our analysis we demonstrated in fact that 485 patients did not exhibit any cardiovascular risk factors or associated vascular diseases. Only 10 (2%) patients had specific atherosclerotic risk factors such as cigarette smoking or mild dyslipidemia. The hallmark symptoms were lower limb pain and claudication observed in 80 (37%) and 66 (30%) patients, respectively. Paraesthesia was observed in 38 (18.5%) patients, weakness in 30 (14%) patients, and monoplegia in 1 (0.5%).

Discussion

We believe that these epidemiological data might have an anatomical explanation. The external iliac artery is in fact a vessel fixed among two points; superiorly, at the level of the iliac bifurcation, and inferiorly at the inguinal ligament. The act of pedalling, the repeated flexion-extension movement, causes a stretching and elongation of the middle portion of the external iliac artery, which, combined with the shear stress forces of the blood flow during exercise, might explain the process of arterial stenosis or occlusion [4]. These anatomo-functional factors are reflected histologically as a remodelling and reactive proliferation of myofibrolasts surrounded by collagen and a substance fundamentally rich in proteoglycans and mucopolysaccharides, resulting in intimal thickening. Arterial endofibrosis, has no inflammatory infiltrate testifying the adaptive process to a mechanical stress rather than to an inflammatory one [5,6]. The ABI index may provide an assessment of disease status and from our analysis, we noticed that in 331 (67%) of the patients there was an ABI drop >0.5, in 123 (24.7%) of the patients there was an ABI between 0.5-0.7 and in 41 (8.2%) patients ABI was normal. In symptomatic young athletes especially cyclists, the diagnostic workup consists of echocolorDoppler which permits in 60% (297 patients) of the cases to demonstrate arterial thickening or kinking both suggesting endofibrosis. Of all the papers reviewed, only 23 papers indicated the exact imaging method used for a total of 38 patients studied. 20 (52%) patients were studied by angiography, 13 (34%) patients underwent CT- SCAN and 5 (13%) patients were studied by MRI. Based on our studies angiography was chosen in the most of the cases. However, at present days, CT scan and MRI are much more accurate than in the past, and we believe that both are the diagnostic tools of choice for this disease. When the disease is diagnosed two therapeutic options are possible: medical or surgical. Medical treatment with anticoagulants and antiplatelets, however, would not seem to be warranted. One (0.2%) patient was treated medically. Furthermore, stopping sports activity should be always recommended to slow the progression of the lesions [2]. The administration of Vit.B1-B6 and folate, if hyperhomocysteinemia is observed, might be considered [2]. The surgical treatment can be either open, laparoscopic or endovascular. In our analysis, 430 surgical procedures in 495 (86%) patients were performed, 411 were open surgeries, (endofibrosectomy, or bypass), and in 5 case the approach was laparoscopic. 13 patients had a percutaneous balloon angioplasty followed by stent placement. Twenty-four works were analyzed to compare the open versus endovascular approach. Only eight of these studies addressed the endovascular approach. Complete information on follow-up at two, six, twelve, and twenty-four months after endovascular procedures was available in only six out of thirteen cases. In two cases, symptomatic restenosis requiring open surgical treatment was detected. However, we observed that the recurrence of the pathology occurred early, after two and six months respectively. We believe that percutaneous transluminal angioplasty with or without stent positioning might be an alternative for selected cases of uncomplicated iliac endofibrosis or an alternative for patients who did not want to undergo surgery. Theoretically, the rate of failure seems to be high. Therefore, we believe that endovascular treatment should be reserved for those patients who intend to stop their sport activities. Endofibrosectomy was the preferred surgical technique (294% to 71% procedures) and a patch angioplasty was associated in 13 (3%) patients. An iliofemoral bypass graft was performed in 117 (29%) patients. In 332 of 441 patients undergo open surgery the median follow-up was 53 months and five-year primary and secondary patency rates either for endofibrosectomy or bypass graft were excellent and similar (reaching 90%). Only in 5/332 (1.5%) patients persist leg pain after surgery. In Therefore, we believe that endofibrosectomy should be preferentially recommended in young patients who want to return to competition whereas bypass graft should be used in adult patients who retired from sport activities and long-standing and complicated lesions [2].

Conclusion

In conclusion we believe that the gold standard is surgical treatment with endofibrosectomy or bypass graft; however, for those patients who do not want surgical treatment, percutaneous transluminal angioplasty with stent positioning might be a possible alternative.

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