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A delayed diagnosis that altered the professional orientation of an athlete with upper limb chronic arterial embolization

**Christos V. Ioannou^{AD}, Alexandros Kafetzakis^{DF}, Christos Kounnos^{BF},
Dimitris Koukoumtzis^F, Emmanuel Tavlas^F, Theodoros Kostas^{AE}**

Department of Vascular Surgery, University of Crete Medical School, Iraklion, Greece

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Summary

Background:

Vascular disorders of the upper extremity in young and physically active patients present a complex and challenging problem for the treating physician. Initial presentation may often be subtle and the consequences of misdiagnosis, delayed diagnosis or mistreatment can be severe.

Case Report:

In this report, we discuss a case of a young woman with chronic upper limb ischemia due to an arterial thoracic outlet syndrome in whom even though symptoms persisted over a number of years during which she frequently sought medical consultation, remained undiagnosed until finally presenting with limb-threatening ischemia. Furthermore, due to this delay, the patient was forced to withdraw from her professional carrier in athletics.

Conclusions:

A thoughtful and through approach combining the history, physical findings, and use of appropriate diagnostic aids will provide the physician and patient with the greatest opportunity for a satisfactory outcome. Furthermore, a delay in definitive treatment may not only cause health deterioration, but may also incur social, economic and occupational consequences.

key words:

thoracic outlet syndrome • cervical rib • post-stenotic aneurysm • critical limb ischemia • misdiagnosis • mistreatment

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Author's address:

Christos V. Ioannou, Department of Vascular Surgery, University of Crete Medical School, PO Box 1352, 711 10, Iraklion, Greece, e-mail: ioannou@med.uoc.gr

BACKGROUND

Sports at all levels place special physical demands on athletes, making them vulnerable to musculoskeletal injury, and may cause a number of sports-related diseases or injuries with coexisting vascular damage [1,2]. Such injuries require immediate medical consultation and are usually easily detected. On the other hand, vascular disorders are low on the list of differential diagnoses, especially when assessing young athletes [3], and often masquerade as other musculoskeletal conditions. This may mislead the unsuspecting medical staff, thus delaying or missing the diagnosis, and often leading to unexpected or even irreversible conditions.

In this report, we discuss a case of a young woman with chronic upper limb ischemia due to an arterial thoracic outlet syndrome in which symptoms persisted over a number of years, and during which the patient was forced to make a significant career change. Even though she frequently sought out medical consultation, her condition remained undiagnosed for years until finally presenting with limb-threatening ischemia.

CASE REPORT

A 26 year-old, non-smoking, female presented with acute deterioration of the right upper limb with severe claudication and weakness. Specifically, she reported a deterioration of her hand claudication which evolved into rest pain with moderate motor and sensory impairment during the previous hours. Detailed history revealed that her right arm claudication began about 6 years prior to this presentation. The patient disclosed that she was attempting to follow a career in professional tennis but was forced to quit due to onset of fatigue in her right arm during practice. She reported seeking medical consultation repeatedly but was always discouraged, with a recommendation to have a number of physiotherapy sessions. This eventually led to her having to quit playing tennis.

Clinical examination revealed a pale, cold arm without palpable pulses distal to the right axillary artery. Her right arm had signs of muscle atrophy. In the supra-clavicle area a pulsatile and tender mass was palpated (Figure 1). Doppler signals were absent in the distal half of the right brachial artery as well as the whole length of the right radial and ulnar arteries. Emergency angiography of the right upper limb revealed an acute occlusion of the brachial artery, indicative of an embolus (Figure 2) and the distal vessels were not visualized. Furthermore, a post-stenotic aneurysm of the right subclavian artery due to a right cervical rib was observed.

Through a supra-clavicle incision, the cervical rib was excised. Prior to aneurysm repair, a thrombo-embolectomy of the limb was performed, clearing the clot up to the brachial bifurcation. An effort to pass the embolectomy catheter into the radial or ulnar arteries was unsuccessful due to chronicity and previous multiple thromboembolic events. Fortunately, there was a patent small branch at the brachial bifurcation which served as an outflow vessel. Postoperatively, the hand clinically improved even though both the ulnar and radial arteries remained occluded.

Further clinical improvement of her claudication was observed at 5-year follow-up, but without complete remission of symptoms.

Figure 1. Clinical evaluation revealed a supraclavicle pulsatile mass. The patient reports having this clinical sign since her early teens.

Figure 2. Angiography revealed a cervical rib (dotted lined arrow) and a post-stenotic aneurysm (solid line arrow) which was site of thrombus generation which in turn caused chronic embolization of the right upper limb leading to the final embolization of the right brachial artery (arrow head).

DISCUSSION

Arterial disorders of the shoulder girdle comprise a diverse group of related problems afflicting the subclavian and axillary arteries [4]. The problems result from a bony anomaly or exaggerated development of the shoulder girdle musculature. Subclavian vein thrombosis is probably the most common vascular problem occurring in athletes [5]. Subclavian artery damage is rare [4], yet subclavian artery thrombosis or aneurysm formation can occur. Vascular sequelae of thoracic outlet syndrome arise as a result of the intermittent but long-term compression of the subclavian artery by a cervical rib, cartilage or fibrous band. Long-term trauma to the subclavian artery leads to degenerative changes within the vessel wall, localized atheroma, and aneurysm formation. Distal thromboembolism may cause acute and chronic upper limb ischaemia, presenting with intense muscular pain on exertion.

The recent increase in sporting activity and associated training has resulted in an increase in the number of patients

presenting with vascular problems [6]. Axillary artery occlusion has been reported in a baseball pitcher [7] and a windsurfer [8], and axillary artery aneurysms have been reported in baseball pitchers [6,9–11] and volleyball players [12]. The mechanism of injury to the axillary vessel is unclear, but there may be damage from the pectoralis minor muscle during repetitive hyperabduction, extension and external rotation of the shoulder [13,14]. Another possibility is damage from bony contact. Duplex scanning of 92 arms revealed direct compression of the artery by the humeral head in 82% of subjects during pitching [15]. It is thought that the sharp downward thrust of the humeral head during a smash in volleyball similarly traumatizes the axillary artery [12].

Our patient had multiple ipsilateral distal arm, forearm and hand arterial occlusions, indicating chronic and repeated embolization. In such scenarios preoperative thrombolysis may have been indicated if the necessary time window for application was available, but this was not performed in our patient because symptoms were gradually intensifying, thus threatening the limb and leading to the operation. Sympathectomy was not performed, but was reserved as a secondary surgical option if limb ischemia did not improve.

Our case is most likely an extreme example of delayed diagnosis and mistreatment and most likely does not represent standard practice. Simultaneously, this case is very dramatic if we consider that even though she was frequently medically examined throughout the 6 years since initial symptom onset, diagnosis was finally obtained only when her limb was threatened. Nehler et al, in a series of 12 patients with upper extremity ischemia from subclavian artery aneurysm caused by bony abnormalities of the thoracic outlet, reported that the average duration of symptoms before correct diagnosis was 7 months (range: 1–36 months) [16]. The previous authors concluded that hand ischemia caused by embolization from a subclavian artery aneurysm occurs in young patients without atherosclerosis and is frequently misdiagnosed as vasospasm [16]. Carpal-tunnel syndrome has also been found to cause a delay in the diagnosis of thoracic outlet syndrome where patients may be subjected to operative carpal-tunnel release before definitive therapy is offered [17].

The number of medical-legal claims has increased greatly in recent years and usually involve claims after surgical interventions [18]. All active medical personnel make an extra effort to avoid finding themselves in the unfortunate position of a medical-legal negligence claim. Previous authors have emphasized the need to inform patients thoroughly about the expectations and risks of any treatment [19]. Furthermore, patients should be re-evaluated after any medical or surgical intervention and if the outcome of a specific therapy is as expected, it may indicate the need to either change treatment or to re-evaluate the initial diagnosis. Our patient sought medical attention and followed various therapies for hypothetical musculoskeletal injuries, without improvement. This was not adequately interpreted during early presentation and after various treatments, and the possibility of another diagnosis was overlooked.

This not only gradually caused a decrease in her tennis-playing performance but led her to eventually present with limb-threatening ischemia. The medical-legal implication of this report highlights the importance of prompt diagnosis and treatment.

CONCLUSIONS

Initial presentation of vascular disorders of the upper extremity in young and physically active patients may often be subtle, and the consequences of misdiagnosis or mistreatment can be severe. A thoughtful and thorough approach combining the history, physical findings, and use of appropriate diagnostic aids will provide the physician and patient with the greatest opportunity for a satisfactory outcome, whereas a delay in definitive treatment may not only cause health deterioration, but may also result in social, economic and occupational consequences.

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