Posterior Circumflex Humeral Artery Aneurysm: Case Report and Systematic Literature Review

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INTRODUCTION
The posterior circumflex humeral artery (PCHA) is an arterial branch originating from the axillary artery supplying the glenohumeral joint and deltoid, minor muscles, and the long head of the triceps muscle.1

PCHA aneurysms are rare, but might lead to ischaemia of the arm, forearm, hand, or fingers. It has been described in overhead athletes such as baseball and volleyball players, where traumatic stretching and compression of the PCHA during repetitive overhead gestures may result in aneurysm formation.2,3 Thrombus embolisation can therefore lead to progressive occlusion of the vessels and consequent ischaemic symptoms.4,5

This is the report of a case of a PCHA aneurysm responsible for digital ischaemia in a young professional volleyball player. A literature review is also presented in an attempt to provide recommendations for PCHA aneurysm diagnosis and management.

REPORT
A 28 year old female volleyball player complained of increasing arm and forearm pain for two weeks associated with unusual right digital cyanosis (Fig. 1). She reported a history of Raynaud’s disease. No palpable pulse was present on the right side on physical examination, whereas all pulses were present on the left. No neurological deficit was noted.

Doppler ultrasound examination (DUS) revealed occlusion of the right brachial, ulnar, and radial arteries. Computed tomography angiography (CTA) in arm abduction and extension as well as magnetic resonance angiography (MRA) confirmed distal embolisation, but no aetiology could be found on these imaging modalities (Fig. 2). The patient underwent an emergency embolectomy through an “S shaped” incision at the level of the elbow and intraarterial fibrinolytics (urokinase) was administered. Postoperative angiography revealed stigmata of embolisation with an incomplete palmar arch and a thin ulnar artery, probably secondary to vasospasm. The patient was discharged on day 1 with oral anticoagulation. Another CTA was performed on day 7 in an attempt to find an aetiological factor. CTA in a neutral position revealed an 18mm diameter PCHA aneurysm (Fig. 3).

It was decided to perform a simple excision of the PCHA aneurysm. The axillary artery was exposed through an axillary approach to avoid harming the shoulder muscles, and the PCHA was identified and ligated (Fig. 4). The PCHA aneurysm was excised after feeding vessel ligation. No revascularisation was required.

The patient was discharged on day 3, and the post-operative course was uneventful. Post-operative DUS revealed patent axillary, ulnar, and radial arteries. The patient was able to play volleyball and returned to competition one month after the surgery.
Although rare, PCHA aneurysms may lead to disastrous complications, such as upper limb ischaemia. Management of a PCHA aneurysm is considered to be a challenge, because it mostly occurs in young professional athletes or workers. PCHA aneurysm is also underdiagnosed, as symptoms are regularly attributed to acute or chronic musculoskeletal injuries. It may result in a career threatening condition, as it can lower performance in healthy and young athletes.

This is the report of a young professional volleyball player presenting with distal embolisation from a thrombosis of the distal part of the axillary artery, the latter related to a PCHA aneurysm. This patient was treated by open repair...
consisting of ligation of the aneurysms without any revascularisation. The diagnosis and management of PCHA aneurysm remains uncertain, because very few cases have been described. A systematic search of the Medline database from 1993 to 2019 by a combined search strategy of MeSH terms (humerus, arteries, aneurysm) was performed.8 All titles and abstracts collected from the search strategy were screened for relevance. The first 20 related items of all relevant articles were scanned for other potentially relevant studies. Full texts of all relevant articles were obtained and reviewed for suitability. The reference lists of each article were scanned for other potentially relevant studies. The systematic search identified 24 English full text studies (Fig. 5), including 11 single case reports, five case series, three reviews corresponding to a total of 52 cases (Table 1).

PCHA aetiology is mostly related to sport. The PCHA travels through a quadrangular space delimited by teres minor muscle superiorly, teres major inferiorly, the humeral neck laterally, and the long head of the triceps muscle medially (Fig. 6). Accordingly, repeated compression between the humeral neck and long head of triceps as well as traumatic stretching of the PCHA during repetitive overhead movements may result in aneurysm formation. Sport related PCHA aneurysms therefore represent 94.2% (49/52) of all PCHA aneurysms reported, and involved sports include volleyball (71.5%; 35/49), baseball (18.4%; 9/49), tennis (4.1%; 2/49), circus trapeze artist (2.0%; 1/49), yoga (2.0%; 1/49), and football (2.0%; 1/49).

Reported symptoms of PCHA aneurysms can vary. PCHA present with digital ischaemia (84.6%; 33/39), arm ischaemia (12.8%; 5/39), or perceptible mass (2.6%; 1/41).

Diagnosis of PCHA aneurysm was made with a single image in the majority of cases (82.6%; 38/46): angiography (43.5%; 20/46), DUS (34.8%; 16/46), MRA (2.2%; 1/46), and CTA (2.2%; 1/46). Two imaging modalities were used in eight cases (17.4%; 8/46).

The optimal management of PCHA aneurysms remains unclear, but surgical resection of the PCHA aneurysm without revascularisation is performed most frequently (60.0%; 18/30), while PCHA resection with revascularisation (20.0%; 6/30), embolisation (13.3%; 4/30), or conservative treatment (6.7%; 2/30) are performed less often. Fibrinolytics (23.3%; 7/30) and surgical embolectomy (20.0%; 6/30) are also described as adjunctive procedures.

Outcomes can be considered as satisfactory because almost all patients were able to return to professional level sport (92.3%; 24/26). Of the two patients who were not able to return to professional level, one continued to experience mild numbness while playing tennis. This patient was treated by resection without revascularisation. The other presented no ischaemic symptoms but suffered from poor sportive performance. He was treated by thrombolysis and surgical resection without revascularisation.

In conclusion, the optimal management for PCHA aneurysms remains unclear, because of the rare occurrence of this pathology. Prompt recognition and diagnosis of PCHA is mandatory, however, to select the best treatment and avoid long term complications. Physicians must be aware of PCHA aneurysm when confronted with young athletes with symptoms of arm or digital ischaemia. Initial imaging should include DUS because it is a non-invasive and affordable imaging modality, but CTA or MRA might also allow an adequate diagnosis. Optimal treatment is unknown, but surgical resection of the PCHA aneurysm either with or without revascularisation, embolisation, or even conservative treatment might be proposed.

Figure 5. Flowchart showing study selection.
| Author        | Date  | Gender | Aetiology                  | Symptoms       | Imaging technique | Treatment                          | Adjunctive procedures | Outcome                  |
|--------------|-------|--------|----------------------------|----------------|------------------|------------------------------------|-----------------------|--------------------------|
| Van De Pol²² | 2018  | MD     | Volleyball player          | Digital ischaemia | DUS MD           | MD                                 | None                  | MD                       |
| Van De Pol¹  | 2017  | M      | Volleyball player          | Digital ischaemia | DUS MD           | None                              | Conservative          | No symptoms              |
| Van De Pol²³ | 2016  | 2F, 11M| Volleyball player          | MD              | DUS MD           | None                              | MD                    | MD                       |
| Kane⁸        | 2013  | M      | Baseball player            | Arm ischaemia   | DUS MD           | Resection with revascularisation   | Surgical embolectomy  | Returned to pitching     |
| Lee⁹         | 2012  | M      | Tennis player              | Arm ischaemia   | Angiography      | Resection without revascularisation | Fibrinolytics         | Returned to competition  |
| Van De Pol⁶  | 2012  | M      | Football player            | Digital ischaemia | Angiography      | Resection without revascularisation | None                  | Returned to competition  |
| Duwayri²¹    | 2011  | M      | Volleyball player          | Digital ischaemia | Angiography      | Resection without revascularisation | Surgical embolectomy  | Returned to professionally pitching |
| Reutter¹⁰    | 2010  | M      | Yoga                       | Arm ischaemia   | CTA              | Resection without revascularisation | Surgical embolectomy  | MD                       |
| Damgaard¹¹   | 2008  | F      | Traumatic (intra-articular injection) | Mass | MRA, CTA | Resection without revascularisation | None                  | No symptoms one year later |
| Seinturier¹² | 2008  | M      | Digital ischaemia          | DUS, Angiography | MD              | Resection without revascularisation | None                  | Poor performance but no more symptoms |
| Baumgarten¹³ | 2007  | M      | Baseball player            | Digital ischaemia | Angiography      | Resection without revascularisation | Fibrinolytics         | Returned to professionally pitching |
| Macintosh¹⁷  | 2006  | F      | Volleyball player          | Digital ischaemia | Angiography      | Resection without revascularisation | Fibrinolytics         | Returned to professionally pitching |
| Tao¹⁴        | 2006  | F      | Volleyball player          | Digital ischaemia | MRA              | Embolisation                       | None                  | Returned to professionally pitching |
| Arko²⁴       | 2003  | M      | Baseball player            | Digital ischaemia | Angiography      | Resection without revascularisation | None                  | No symptoms              |
| Vlychou¹⁵    | 2000  | M      | Volleyball player          | Digital ischaemia | Doppler, Angiography | Embolisation                       | Surgical embolectomy  | MD                       |
| Ikezawa⁷     | 2000  | M      | Tennis player              | Digital ischaemia | Angiography      | Resection without revascularisation | Surgical embolectomy  | Mild numbness on the second and third fingers when playing tennis |
| Schneider¹⁶  | 1999  | M      | Baseball player            | Digital ischaemia | Angiography      | Resection with revascularisation   | Fibrinolytics         | Returned to professionally pitching |
| Reekers¹⁸    | 1998  | M      | Professional circus trapeze artist | Digital ischaemia | Angiography      | MD                                 | None                  | MD                       |

Table 1. Posterior circumflex humeral artery aneurysms: literature review
CONFLICTS OF INTEREST
None.

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None.

APPENDIX A. SUPPLEMENTARY DATA
Supplementary data to this article can be found online at https://doi.org/10.1016/j.ejvssr.2019.07.001.

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