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### **REVIEW**

# Popliteal Artery Entrapment Syndrome in Children: Experience With Four Cases of Acute Ischaemia and Review of the Literature

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#### WHAT THIS STUDY ADDS

Popliteal artery entrapment syndrome (PAES) is a well known entity in adults and has been widely documented in the literature. This anatomical abnormality is uncommon in paediatric practice and is often an unheralded diagnosis, the disease often being discovered at the stage of complications. In this report clinical and surgical experience of PAES in children with acute ischaemia is detailed. In addition, data were analysed from the literature with emphasis on the clinical severity on admission. The various diagnostic strategies and the outcome of treatment in paediatric patients were assessed. This paper gives interesting information about an extensive review of PAES cases in children not reported before.

**Objective:** Popliteal artery entrapment syndrome (PAES) is an uncommon anatomical anomaly, frequently described in adults. The most common symptom is claudication. Acute limb ischaemia (ALI) in children is rare, but it may evolve and lead to limb loss or lifelong complications. Clinical and surgical experience of PAES in children is reported. Data from the literature are analysed in order to assess the severity of this disease and to identify the factors characterising the diagnosis and the outcome of treatment in paediatric patients.

Methods: Four children (aged 7—16 years) were referred with ALI due to PAES. Among the 439 articles reporting cases of PAES, 55 patients under 18 years of age were the focus. The PAES cases were classified according to the Love and Whelan classification modified by Rich.

Results: Data from 79 children (106 limbs, 27 bilateral PAES) were collected and analysed. Type I PAES was present in 41 (39%), Type II in 23 (22%), Type III in 24 (23%), Type IV in 12 (11%), and Type V in two (2%) limbs. A functional PAES was present in one patient bilaterally. In two cases, the type of PAES was not reported. Claudication occurred in 68 cases (64%), and ALI in 19 (18%). In 60 cases (57%), revascularisation with or without myotomy was required; myotomy alone was performed in 41 cases (39%).

**Conclusions:** Symptomatic PAES in children should be considered a severe condition requiring urgent investigation in order to avoid any delays in the treatment. Early diagnosis and treatment are essential to prevent serious complications. The long-term outcomes of surgical treatment with the correction of the anatomical anomaly and vascular reconstruction are satisfactory with a low complication rate.

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#### **INTRODUCTION**

Popliteal artery entrapment was first described by Stuart in 1879<sup>1</sup> as an abnormal anatomical relationship between the popliteal artery and local musculo-tendinous structures caused by an anomaly of embryological development. The term popliteal artery entrapment syndrome (PAES) was introduced by Love and Whelan in 1965.<sup>2</sup> PAES is caused by

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compression of the popliteal artery by the surrounding musculo-tendinous structures.

The most common symptom of PAES is lower leg pain with calf claudication. This mostly occurs in young adults (30—40 years) with well developed muscles, such as soldiers and athletes.<sup>3,4</sup> However a number of reports have described cases of children presenting with acute lower limb ischaemia caused by PAES.<sup>5,6</sup>

Acute limb ischaemia (ALI) in children is rare, but it may lead to limb loss or lifelong complications. Several well known factors, including embolic syndromes, thrombophilia, and hypercoagulable states, can cause ALI. The diagnosis in children is often delayed since the first symptoms, including pain and claudication, are not reported by paediatric patients, hampering the possibility of early surgical treatments.

In this study, the aim was to report the experience of PAES in children and to analyse data from the literature in order to assess the severity of this disease and to determine the factors characterising the diagnosis and outcome of treatment in paediatric patients.

#### **METHODS**

Four children (aged 8—16 years) were referred to hospital with ALI due to PAES between 2004 and 2009.

For the literature review, the following search strategy on the Medline database for popliteal entrapment syndrome was used: ("popliteal artery" [MeSH Terms] OR ("popliteal" [All Fields] AND "artery" [All Fields]) OR "popliteal artery" [All Fields]) AND entrapment [All Fields]. Altogether, 436 articles were identified. Literature research results were collected until March 2016. All papers that included patients under 18 years of age were selected for a detailed review (Fig. 1). Articles were excluded if the age of the treated patients was not mentioned in the paper or if the cases were not described. Three further articles were found from the reference lists of the selected papers. There were patients with PAES who were under 18 years old in 55

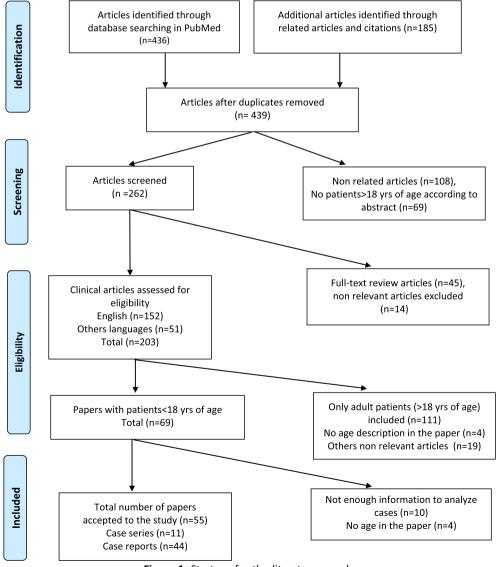


Figure 1. Strategy for the literature search.

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