

# Venous aneurysm as a clinical problem. General and local complications

## Tętniak żylny jako problem kliniczny. Powikłania ogólne i miejscowe

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### Abstract

Venous system aneurysms appear relatively rarely and are diagnosed even less frequently, especially if asymptomatic. Asymptomatic aneurysms are detected during duplex-Doppler studies. Symptomatic aneurysms are often life-threatening.

The first part of this paper presents a literature review of this rare disease, and the second part presents a the material of the cohort of 5 patients underwent surgical treatment due to venous aneurysm and its complications.

There is no clear standard of treatment for venous aneurysms. The main reasons for this are relatively low epidemiology, diversity of locations, difficulty in establishing primary and secondary aetiologies, anatomical structure, and the coexistence of concomitant diseases. Given the numerous uncertainties, several factors should be considered when deciding the next steps in treatment.

**Key words:** venous aneurysm; clinical symptoms; rupture of a venous aneurysm; differential diagnosis; therapy

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### Streszczenie

Tętniaki układu żylnego pojawiają się stosunkowo rzadko i są diagnozowane jeszcze rzadziej, zwłaszcza jeśli są bezobjawowe. Bezobjawowe tętniaki są wykrywane podczas badań ultrasonograficznych. Objawowe tętniaki często zagrażają życiu.

W pierwszej części artykułu przedstawiono przegląd literatury na temat tej rzadkiej choroby, a w drugiej części przedstawiono materiał i dane kliniczne 5 chorych z tętniakiem w układzie żylnym poddanych leczeniu operacyjnemu.

Nie istnieje jednoznaczny standard leczenia tętniaków żylnych. Głównymi przyczynami są stosunkowo rzadkie występowanie, różnorodność lokalizacji, trudności w ustaleniu etiologii pierwotnej i wtórnej, struktura anatomiczna oraz obecność chorób współistniejących. Biorąc pod uwagę liczne wątpliwości przy podejmowaniu decyzji o kolejnych krokach leczenia, należy wziąć pod uwagę szereg czynników.

**Słowa kluczowe:** tętniak żylny; objawy kliniczne; pęknięcie tętniaka żylnego; diagnostyka różnicowa; terapia

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## Introduction

Venous aneurysm belongs to a group of rare pathologies that have only recently been described and relates to both the vascular and venous system. The first description was presented by May and Nissel [1] in 1968. Above all, there is no clear statement on the extent to which the location, size, and shape of the aneurysm determine the risk. Currently, there are practically no collective statistics about venous aneurysms.

Since this first report, over 100 cases of venous aneurysms located mainly in the area of internal jugular veins, the saphenous vein, the inferior vena cava, the superficial femoral vein, and the popliteal veins have been presented [2]. The current literature indicates that aneurysms are neither sex nor age-dependent [3]. Similarly, the aetiology of the condition has not been clarified. It is assumed that the cause may be congenital hypoplasia of the aneurysm-altered vein wall. The few existing publications on histological studies of surgical preparations have shown no post-traumatic, rheumatoid or post-inflammatory changes [4]. The course of the disease usually involves the possibility of pulmonary embolism; the condition that usually leads to a correct diagnosis. In all cases of unexplained pulmonary embolisms, especially recurrent ones, the possibility of dissemination from the venous aneurysm should be considered. A correct diagnosis is possible after detailed examinations [5]. Physical examination of the accessible vein sections sometimes reveals a weakly movable soft tumour (rarely painful). In the literature, a popliteal venous aneurysm is the most thoroughly described aneurysm. It has broader symptomatology, causing periodic swelling of the lower leg, pain, and development of signs of post-thrombotic syndrome. It is the fourth most frequent venous aneurysm, following jugular vein and splenic vein aneurysms. The differential diagnoses should also consider a Baker's cyst. Regrettably, a popliteal venous aneurysm is connected with a high mortality rate due to pulmonary embolisms, which affect over 70% of patients. 88% of popliteal venous aneurysm cases are diagnosed based on unexplained, recurrent pulmonary embolisms [6]. The article summarizing all world literature regarding this topic stated that 105 cases were described [7]. According to Berquist, the quality of individual reports is varying. Some of them do not contain all of the necessary information, especially regarding further procedures, both in the context of follow-up time and outcome. Symptomatology of the analysed 1–5 cases patients was as follows: chest symptoms (46 cases) and suspicion of pulmonary embolism, deep vein thrombosis (6 cases), with local symptoms, palpable masses (38 cases), venous insufficiency (14), arterial symptoms (4 cases), and asymptomatic (4 cases). In addition to the 105 patients, the authors added 17 case reports published by Voirin et al. and 25 cases reported by Sessa et al. [2, 8]. The authors of the analysis discussed the difficulty with the analysis of these added reports (according to [7]). Based on publications, the basic and most reliable diagnostic test was phlebography, carried out in 2/3 of

cases. Much less frequently, the diagnosis was based on an ultrasound scan, angio-MRI, or angio-CT. Predominantly, the basic treatment was based on surgical procedures ranging from ligation of the aneurysm (through aneurysmorrhaphy) to revascularization procedures with the interposition in of the PTFE prosthesis [9]. Apart from the basic symptoms mentioned above, several other, unexpected symptoms appeared. Three young patients demonstrated cerebral vascular embolism resulting from a paradoxical (crossed) embolism through an oval unsealed opening. A combination of symptoms in the form of pulmonary artery embolism, left popliteal fossa resistance, and intermittent claudication caused by compression of the popliteal vein aneurysm from the pressure of the popliteal artery aneurysm were described.

A separate clinical problem constitutes a visceral vein aneurysm. In a published paper based on 93 reports, descriptions of 198 venous aneurysms in 176 patients were found and are considered to be extremely rare clinical problems [10]. Aneurysms were mostly within the portal system (87 of 93 reports, 170 of 176 patients, 191 of 198 aneurysms). Individual cases of renal and mesenteric vein changes were also described. The existence of an aneurysm within the visceral vein system was associated with abdominal pain in 44.7% of patients, gastrointestinal bleeding in 7.3%, and asymptomatic presentation in 38.2% of patients. In the 1995 retrospective evaluation of Calligaro et al., four cases of the jugular venous aneurysm and one case of basilica vein aneurysm were diagnosed. Three aneurysms of the internal jugular vein were followed up for four years in colour duplex ultrasound and no complications occurred. According to these authors, aneurysms of other parts of the venous system are a much greater problem [11, 12]. In a group of 32 patients with abdominal venous aneurysms (18 portal, 7 inferior vena cava, 4 superior mesenteric veins, 2 splenic, 1 internal iliac), 13 (41%) had serious complications, including 5 deaths. Out of 31 patients with deep venous aneurysms of the extremity (29 popliteal, 2 common femoral), 22 (71%) had deep vein thrombosis or pulmonary embolism. In 17 cases of deep vein thrombosis or pulmonary embolism, patients were treated with anticoagulants or underwent surgery. However, other venous aneurysms should be removed only if they are symptomatic, enlarging, or disfiguring [13, 14].

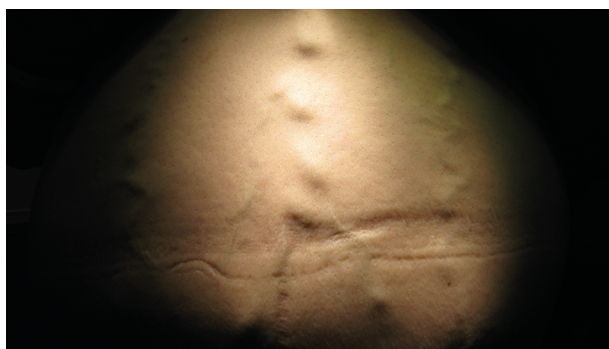
## Material and methods

Five patients with a diagnosis of venous aneurysm were diagnosed and treated in the study department. The diagnosis was typically made based on symptoms, ultrasound, CT, and MRI results. Each case was unique with varying dynamics of their condition and state. For this reason, the procedures used were different with the use of both conservative and surgical treatment.

The first case of interest was a 68-year-old patient. An ultrasound revealed a tumour of the retroperitoneal space measuring 65 mm in diameter and 120 mm in length. CT scan showed that the tumour was a wide inferior

vena cava filled with thrombi and no other pathology. An angio-CT scan demonstrated occlusion of the V. cava inferior and collateral circulation through the spinal veins. Physical examination revealed the presence of the extensive collateral venous circulation on the abdomen as well as large varicose veins in both lower limbs. The biopsy material was collected endovascularly. No other cells were found in the examination — only a clot. The patient was suitable for conservative treatment with anticoagulants due to well-developed collateral circulation, mild clinical symptoms, and the surgery carried significant risks (Fig. 1, 2).

The second case represented an example of a pulmonary embolism caused by a venous aneurysm.



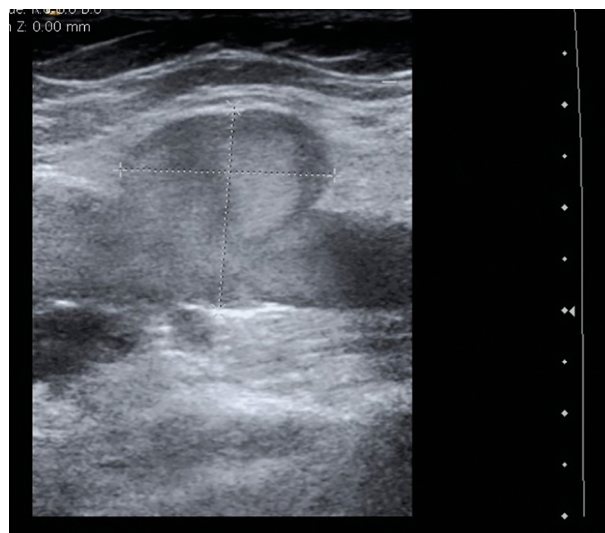
**Figure 1.** The visible collateral venous circulation in the abdomen due to caval vein occlusion related to the venous aneurysm



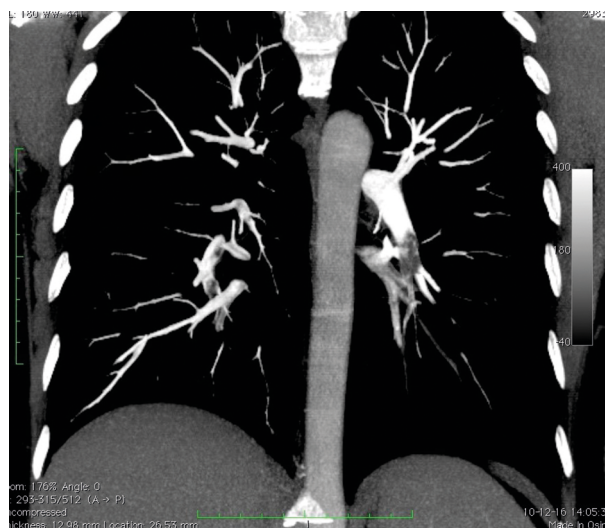
**Figure 2.** An thrombosed giant aneurysm of the inferior vena cava

A 54-year-old female patient was admitted due to a tumour on the medial side of the right arm. On physical examination, reddening of the skin was observed over the palpable longitudinal tumour and the patient had pain on palpation. Ultrasonography revealed a venous aneurysm with thrombotic (both old and fresh) material present in the absence of a suspected arteriovenous fistula (Fig. 3).

In addition, the patient complained of a deterioration in cardiopulmonary function that appeared suddenly six months earlier and was associated with haemoptysis. A similar incident occurred a few days before admission to the hospital. Further diagnostics with perfusion and ventilation scintigraphy were performed, allowing for the diagnosis of peripheral pulmonary embolism. In these circumstances to clarify the diagnosis, a lung angio-CT was also performed (Fig. 4) — showing the presence of multiple pulmonary emboli.



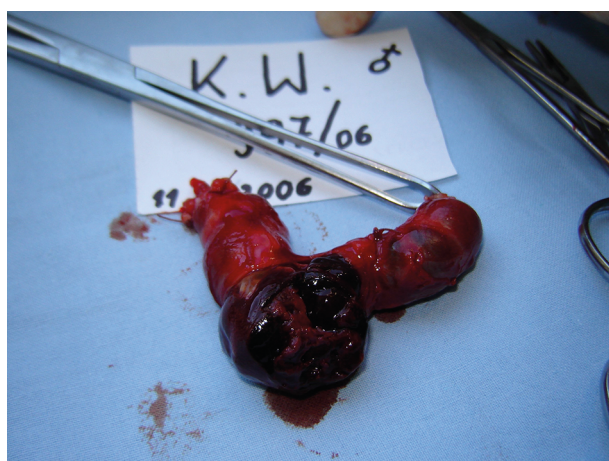
**Figure 3.** Ultrasound image — brachial vein venous aneurysm in the right upper limb



**Figure 4.** Image of multiple pulmonary emboli related to the upper limb venous aneurysm presence



**Figure 5. Intra-operative image — brachial vein venous aneurysm**



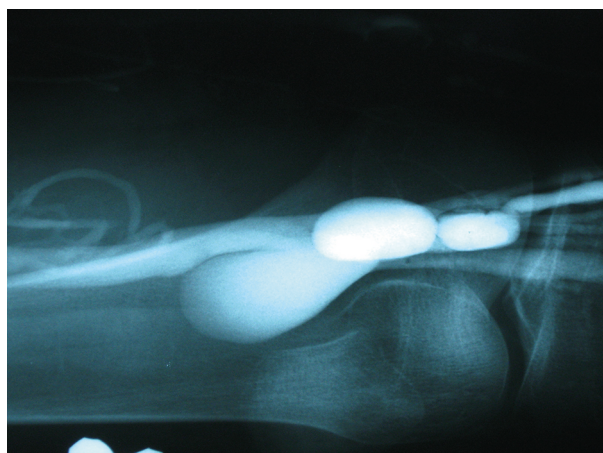
**Figure 6. Resected brachial vein venous aneurysm**

The patient was eligible for surgical treatment and the lesion was excised. An intraoperative picture and cross-section of the resected aneurysm are shown in the following figures (Figs. 5, 6). After resection of the aneurysm, symptoms of venous embolism did not recur at the 24-month follow-up. Anticoagulation medications were administered for 6 months (orally).

Histopathological examination of the brachial vein showed degeneration of smooth muscle cells and collagen fibres and local hyperplasia.

The two other cases involved young patients with venous aneurysms demonstrated by phlebography, however, they did not contain thromboses (Fig. 7). The patients were asymptomatic, without thromboses, and did not have pulmonary embolism symptoms. It was decided to undertake conservative treatment with compression therapy and frequent ultrasound follow-up. At the five- and seven-year follow-up, patients were asymptomatic.

The last patient description concerns a fit man (36 years old) which was admitted due to the swelling



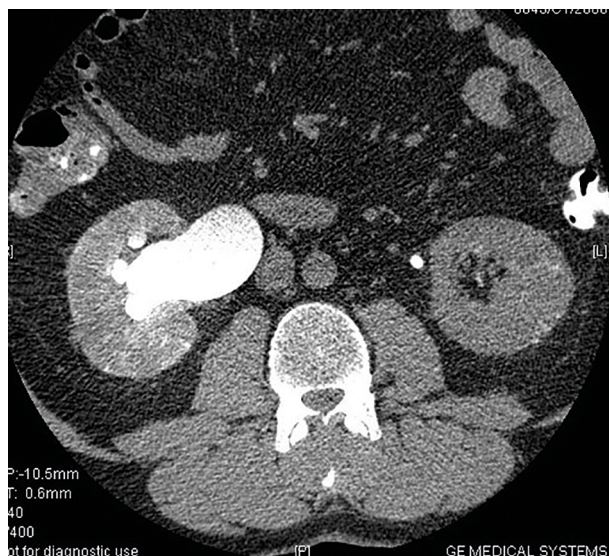
**Figure 7. Multiple aneurysms of the popliteal vein**

of the right leg that had gradually grown over the past 14 days. The temporary swelling had occurred before and the patient associated it with physical effort. Simultaneously with the swelling, the patient was experiencing increasing pain in the right lumbar region, which was also initially transitory in nature. Recently, the pain, as well as the swelling of the lower limb, had become constant. Due to the unclear nature of the condition, he was treated by the general practitioner with anti-inflammatory drugs with no effect. On the day of admission to the hospital, a physical examination revealed lower abdominal soreness and guarding with no peritoneal symptoms. In the initial blood count, apart from lowered morphology values (Hb — 10 g/l, Ht 30%), all other parameters were normal, including leukocytes, urea, and creatinine. The abdominal ultrasound scan showed a large haematoma in the pelvis minor with venous flow. Additionally, it revealed changes within the urinary system in the form of a large right-sided hydronephrotic kidney, and ureteral dilatation extending to the level of the iliac tuberosity (compression) (Fig. 8).

CT and angio-CT scans showed the presence of a large haematoma within the lesser pelvis. This exerted significant pressure on the ureter and led to the subsequent hydronephrosis of the right kidney. Under these circumstances, rupture of a venous aneurysm was suspected (Fig. 9). Moreover, there was suspicion of compensation symptoms due to the chronic venous insufficiency of the right lower limb caused by compression on the external iliac vein with no thrombotic changes. Detailed medical history did not reveal any pertinent information including past medical history from his family members.

Immediately after the completion of the diagnostic procedures and the decision to pursue surgical treatment, the patient's condition deteriorated rapidly and symptoms of haemorrhagic shock occurred.

The patient was operated on immediately, with simultaneous implantation of a temporary filter into the inferior vena cava. After the evacuation of numerous blood clots (volume ca. 1000 ml), it was determined that the source of



**Figure 8. Hydronephrosis of the right kidney**

fresh bleeding was the ruptured baggy aneurysm of the internal iliac vein that was “sitting” on the common iliac artery bifurcation. The main surgical procedure involved performing aneurysmorrhaphy with the restoration of the physiological venous continuity of the right venous axis. A further section of the internal iliac vein was ligated. In the postoperative period, venous stasis was found to have subsided. In the absence of symptoms of pulmonary embolism in clinical, laboratory, and cardiac ultrasound examinations, the filter from the inferior vena cava was removed. The hydronephrosis of the right kidney, which was previously observed, had also receded. After a two-year follow-up period, the patient is symptom-free. Histological examination showed pathological degeneration of smooth muscle cells, collagen fibres, and local lesions related to the internal iliac vein hyperplasia.

## Results

According to the reports of the patients treated in the authors’ centre, the presence of a venous aneurysm in five cases was analysed. In three of the cases, patients were acceptable for conservative treatment, and in two cases surgery was necessary. The diagnosis in all five cases was made based on symptoms, ultrasound, CT, and MRI results. The patient presenting with the large, ruptured aneurysm of the internal iliac vein was diagnostically complex, due to the speed at which the symptoms were worsening, and eventually was forced to proceed to an emergency procedure. None of the patients died perioperatively.

The course of the disease is often associated with the possibility of pulmonary embolism, a condition usually leading to death or the final diagnosis. In all cases



**Figure 9. Visible aneurysm of the right internal iliac vein**

of unexplained pulmonary embolism, the possibility of a venous aneurysm should be considered. Correct diagnosis is not easy and is only possible following thorough examinations, including ultrasound, CT, and MRI. Physical examination of the accessible sections of the vein sometimes reveals an immovable, typically painless, soft tumour.

## Discussion and conclusion

There is no clear standard of treatment for venous aneurysms. The main reasons for this are relatively low epidemiology, diversity of locations, difficulty in establishing primary and secondary aetiologies, anatomical structure, and coexistence of concomitant diseases [15–17]. Despite numerous uncertainties, several issues should be considered first when deciding the next steps. Undoubtedly, the very structure, location, and size of the aneurysm are of importance in the decision. Equally as important is the presence or lack of thrombotic material. All of these factors should be considered in assessing the risk of complications such as venous hypertension, venous insufficiency, and pulmonary embolism.

In the case of venous aneurysms, the decision about the treatment method is always difficult and requires a lot of experience. According to KA Teter, “Management of these lesions is determined largely by location and the potential morbidity and mortality of the untreated aneurysms vs surgery and endovascular techniques continue to lack a defined role in their management, and the standard of care remains open repair when indicated” [18].

## Conflict of interest

None declared

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