Radiology Case Reports 16 (2021) 1284–1287

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journal homepage: www.elsevier.com/locate/radcr

Case report

Transcatheter arterial embolization for a symptomatic Tarlov cyst with hemorrhage due to an underlying arteriovenous fistula ☆,☆☆

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ARTICLE INFO

Article history:
Received 26 January 2021
Revised 23 February 2021
Accepted 24 February 2021

Keywords:
Tarlov cyst
Hemorrhage
Arteriovenous fistula
Transcatheter arterial embolization

ABSTRACT

Symptomatic sacral perineural cysts (Tarlov cysts) accompanied by intra-cyst hemorrhage are rare. The treatment strategies have not been established. We report a 57-year-old woman with severe back pain due to a Tarlov cyst accompanying intracyst hemorrhage. Computed tomography angiography revealed an arteriovenous fistula (AVF) at the area surrounding the cyst. The patient underwent transcatheter arterial embolization for the AVF. Thereafter, the hematoma and cyst decreased in size, and clinical symptoms markedly improved with no additional surgery. Transcatheter arterial embolization may be an effective alternative to surgery for Tarlov cysts with vascular disease, including AVF.

Introduction

Sacral perineural cysts (Tarlov cysts) are observed in the extradural segment between the perineurium and endoneurium. The lesions are incidentally found in 1%-4.6% of cases by lumbosacral magnetic resonance imaging (MRI) [1,2]. Most Tarlov cysts are asymptomatic and do not require specific intervention, whereas approximately 13%-22% are symptomatic and require treatment [2,3]. Symptomatic Tarlov cysts accompanied by intra-cyst hemorrhage are markedly rare and the majority were reported to be associated with subarachnoid hemorrhage (SAH). The treatment strategies for symptomatic Tarlov cyst have not been established. We report a symptomatic Tarlov cyst accompanied by intra-cyst hemorrhage due to an arteriovenous fistula (AVF), which was successfully treated by transcatheter arterial embolization (TAE), resulting in a good clinical outcome.

Case presentation

A 57-year-old woman was admitted to our hospital with severe back pain and right lower limb pain. She had a history of Tarlov cyst, angina pectoris, and neurofibromatosis type 1

☆ Competing Interests: The authors declare that they have no conflict of interest.
☆☆ Funding: This work received no specific grant from any funding agency in the public.

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https://doi.org/10.1016/j.radcr.2021.02.056

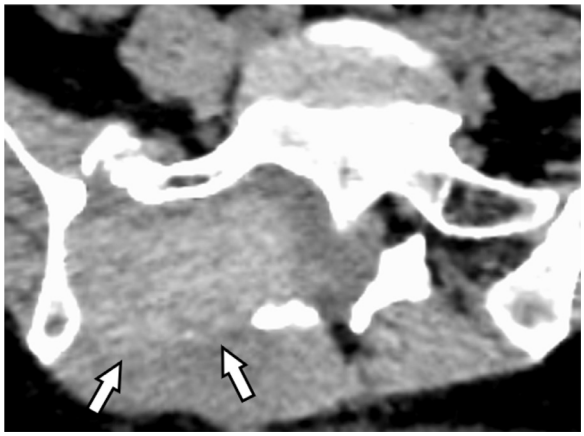

Fig. 1A – Plain CT showed a hyperdense mass (arrows) corresponding to Tarlov cyst between the right sacral foramen and spinalis muscles. The right sacral foramen was dilated by the lesion at the level of the first and second nerves.

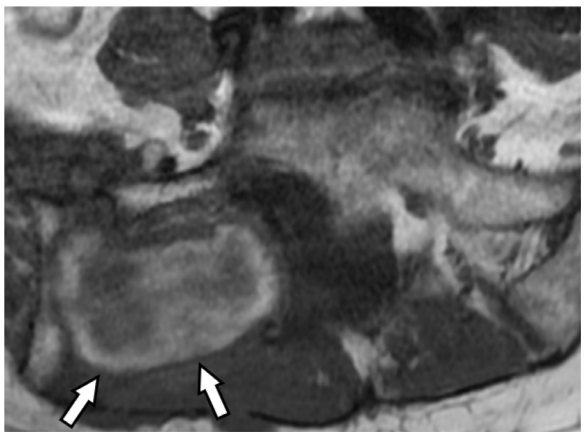

Fig. 1B – T1-weighted magnetic resonance imaging (MRI) showed the periphery of the tumor (arrows) had high-intensity signal and the inside had low-intensity signal, suggestive of hematoma in the Tarlov cyst.

(NF1). Plain computed tomography (CT) revealed dilatation of the right sacral foramen at the level of the first and second nerves, and a hyperdense mass from the right sacral foramen to spinalis muscle (Fig. 1A). Magnetic resonance imaging revealed the mass originating from the first and second right sacral nerve roots (Fig. 1B). The periphery of the mass had high-intensity signal and the inside had low-intensity signal on T1-weighted image, suggestive of hematoma in the Tarlov cyst. CT angiography revealed an AVF between the right lateral sacral artery and right internal iliac veins at the area surrounding the hemorrhage (Fig. 1C). There were no signs of active bleeding or pseudoaneurysm. Subsequently, surgical removal of the hematoma was performed, but the operation failed due to massive hemorrhage. As a result, no reduction in hematoma was obtained. Pre-operative TAE was then attempted to minimalize bleeding during the next surgery.

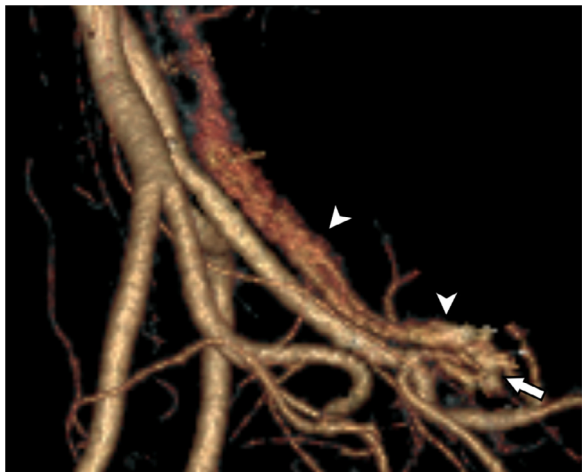

Fig. 1C – CT angiography showed early venous drainage into the right internal iliac vein (arrowheads) caused by the arteriovenous fistula. The main feeding artery was the right lateral sacral artery (arrow).

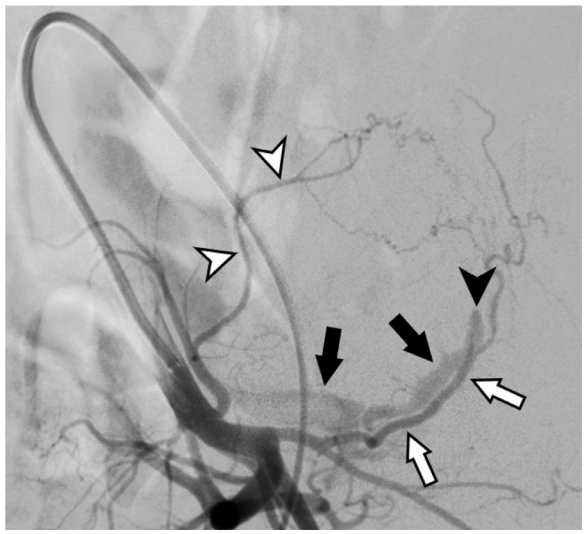

Fig. 2A – Right internal iliac arteriography before embolization showed an arteriovenous fistula (black arrowhead), and the right lateral sacral artery (white arrows) and right iliolumbar artery (white arrowheads) directly communicating with branches of the right internal iliac vein (black arrows).

A 6-Fr guiding sheath (Parent Plus, Medikit, Tokyo, Japan) was percutaneously inserted from the left femoral artery under local anesthesia. Right internal iliac arteriography revealed that the right lateral sacral and iliolumbar arteries directly communicated with the internal iliac vein with early venous drainage, suggesting an AVF with high flow (Fig. 2A). A microcatheter (Tellus, Asahi Intec., Aichi, Japan) was coaxially advanced near the fistula. The feeding arteries were completely embolized using micro coils. No further early venous drainage was observed (Fig. 2B). No adverse events occurred during or after the procedure.

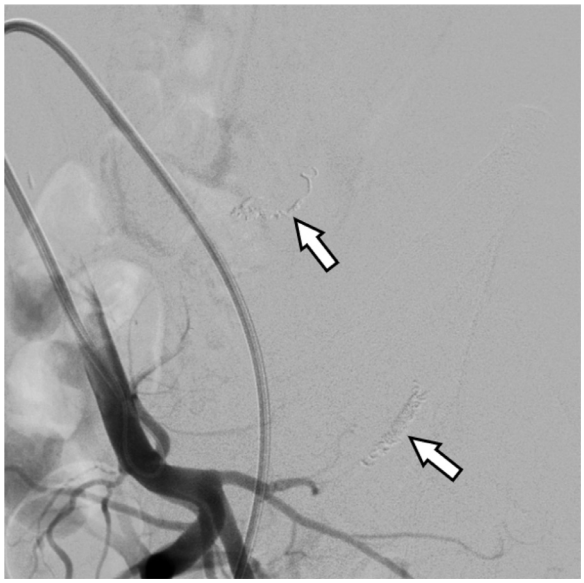

Fig. 2B – Right internal iliac arteriography after embolization showed no early venous drainage. Arrows indicate embolic metallic coils.

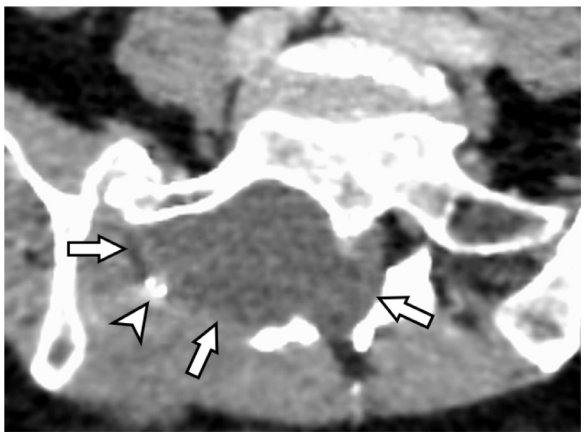

Fig. 3 – Contrast-enhanced CT 1 month after embolization showed the Tarlov cyst (arrows) and intracyst hematoma markedly decreased in size. Note that embolic metallic coils (arrowhead) placed in the right lateral sacral artery were located in the area surrounding the cyst.

As her symptoms improved immediately after TAE, the planned surgery was canceled. The patient was discharged 5 days later. CT one month after embolization demonstrated that the hematoma and cyst markedly decreased in size (Fig. 3). At the 20-month follow-up, the patient had marked improvement in clinical symptoms, such as back pain, and was free from the administered analgesic.

Discussion

The present case included two unique findings. First, the patient had symptomatic Tarlov cysts accompanied by intra-cyst hemorrhage secondary to AVF, which was not previously reported. Second, TAE was an effective alternative to surgery, resulting in hemostasis, good reduction of the hematoma, and marked improvement in clinical symptoms.

Tarlov cysts arise between the arachnoid covering the nerve root and the outer layer of its pia cover [4]. The cysts may increase in size due to hydrodynamic and pulsatile forces of cerebrospinal fluid (CSF) infiltration into cysts and disturbance of the exit, termed the ball-valve mechanism [5]. The cysts often communicate with CSF flow. When the cyst fills with fluid, such as CSF or blood, the adjacent neural or body structure is compressed and pain develops. Pain after hemorrhage in the cysts is associated with SAH after rupture of a cerebral aneurysm [6]. In the present case, although SAH secondary to rupture of a cerebral aneurysm was not identified, the intra-cyst hematoma likely expanded the lesion, leading to clinical symptoms.

Of note, the present case included an AVF in the Tarlov cyst. The AVF may have been associated with the following factors: the current patient had NF1, which can cause fragility of the vessel wall and marked vascularity of the surrounding neurofibromatous tissue; arterial aneurysm dissection and AVF, as previously reported [7]. In addition, the patient frequently underwent nerve block for persistent pain. Therefore, the background, including vasculopathy from NF1 and frequent nerve block, were considered to be associated with the presence of AVF. Furthermore, the patient received antiplatelet therapy for angina pectoris. This may have played a role in the intracyst bleeding, probably secondary to microfracture of the AVF.

The present case suggested that the intra-cyst hematoma was associated with underlying AVF. Indeed, CT angiography demonstrated that the fistula was located at the area surrounding the Tarlov cyst. Clinical symptoms improved immediately after embolization of the fistula, and the hematoma and cyst subsequently decreased in size.

The treatment strategies for symptomatic Tarlov cysts have not been established. Many treatments for symptomatic Tarlov cysts, including microsurgical cyst excision, anti-inflammatory medication, selective nerve root block, cyst aspiration, percutaneous injection of fibrin sealant, cyst-peritoneal shunts, and conservative treatment, have been reported [8,9]. To our knowledge, the current case is the first successful performance of TAE for a symptomatic Tarlov cyst with intra-cyst hemorrhage secondary to an AVF.

TAE can be effective for vascular diseases such as AVF. This minimally invasive therapy provides occlusion of the fistula through a catheter super-selectively inserted into the feeding arteries of the AVF. In the present case, the main clinical symptoms markedly improved immediately after embolization, with a reduction in the volume of the cyst and hematoma. Thus, TAE helped to treat the symptomatic Tarlov cyst secondary to the AVF.

In conclusion, when hemorrhage in symptomatic Tarlov cysts is observed, the presence of underlying vascular lesions, such as AVF, should be investigated by CT angiography. In such cases with AVF, TAE may be a promising alternative to surgery.

Patient consent

Written informed consent was obtained from the patient for publication of this manuscript.

Ethical approval

This article was approved by the institutional review board.

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