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Intradural Perimedullary Arteriovenous Malformations: Vascular Structure and Surgical Treatment

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Abstract

Background: Intradural perimedullary arteriovenous malformations arisen on the surface of the spinal cord and described mostly as fistulas are relatively rare lesions. Recently it was suggested that they probably are a mixed group of malformations located on the surface of spinal cord.

Methods: The radiologic and operative features of perimedullary arteriovenous malformations were analyzed in 42 consecutive cases. The patients were treated by endovascular, surgical or combined occlusion\resection. Surgical and endovascular treatment results were carefully analysed and evaluated.

Results: Perimedullary vascular malformations were subclassified into 4 groups: 1) cervical intradural perimedullary fistulas; 2) intradural perimedullary arteriovenous fistulas in thoracic region; 3) perimedullary arteriovenous fistulas of the spinal cord cone; and 4) perimedullary arteriovenous malformations. The prominent neurological improvement after treatment was observed in 9 patients. Partial regress was demonstrated in 21 patients, and in 9 patients the neurological symptoms remained unchanged. Clinical deterioration was observed in 3 patients.

Conclusions: In perimedulary arteriovenous malformations, a surgical strategy should be based on the knowledge of changes in physiological parameters such as blood flow and of the individual anatomy of malformation. It was always essential to disconnect the distal portion of feeding vessel near a place where they empty into the draining veins.

Keywords: Intradural Perimedullary Vascular Malformations; Surgical treatment

Introduction

The current classification system of spinal arteriovenous malformations (AVM) comprises 4 types based on the angiographic features and pathophysiology of the lesions. Type I spinal AVMs are the dural AVMs in which the radicular arterial feeder forms a fistula in the dural root sleeve. Type II spinal AVMs are the glomus lesions and consist of a compact, intramedullary nidus. Type III spinal AVMs are also known as juvenile type and are mostly intramedullar lesions that may also have an extramedullary and even an extraspinal extension [1]. Intradural perimedullary AVMs (type IV), the most poorly investigated lesions, are localized on the surface of a spinal cord and they do not penetrate the spinal cord and do not fall outside the limits of dura [2]. Type IV AVMs are traditionally considered as true arteriovenous fistulas [3]. These AVMs often happen to be a challenge to neurosurgeons due to complicated varieties of blood supply including feeding and draining spinal vessels with high blood flow. So far the results of the surgical treatment remain unsatisfactory [2].

We analysed our ten-year experience in the surgical treatment of patients with spinal AVMs localized intradurally, perimedullary and which can not be classified differently except for as type IV spinal vascular malformations. We summarized the data about these AVMs, their localization, vascular anatomy and effects of surgical treatment. The main aim of the study was to find out whether the effects of endovascular or surgical treatment depend on the localisation and vascular anatomy of AVM.

Materials and Methods

Forty-two patients with intradural perimedullary vascular malformations were retrospectively studied. Table 1 demonstrates localisation, anatomy and blood flow of AVMs. From all intradural perimedullary vascular malformations we identified 34 intradural perimedullary arteriovenous fistulas (IPAVF's) and 8 intradural perimedullary arteriovenous malformations (IPAVMs). All IPAVMs were localized in a thoracic region of the spinal cord. In contrast, the IPAVF's had a number of different localizations. In cervical region all the IPAVF's fistulas were feeding from radiculomedullary arteries beginning itself from vertebral artery (n = 9). In 18 patients, IPAVF's were localized in the thoracic region of the spinal cord; they were feeding either from the anterior and posterior spinal arteries or from the radiculomedullary arteries. In 7 cases, the conus medullaris IPAVF's were diagnosed (Table 1).

Based on the blood flow intensity, all perimedullary vascular malformations were subdivided into 3 standard types: A, B, and C.

The diagnosis of IPAVF and IPAVM was made based on the results of MRI and spinal or vertebral angiography.

The selection of optimal management of spinal IPAVF's, IPAVMs requires analysis of their localisation, vascular anatomy, and blood flow with a review of empirical data obtained through various treatment strategies. Endovascular treatment, open microsurgical procedures or a combination of both approaches were used (Table 2). The surgical interventions were directed at switching off of the feeding vessels near the fistulae or AVM, or resection of IPAVM. Occasionally, we resected tortuous draining veins of IPAVM, IPAVF.

Results

Localisation and vascular anatomy

We studied the vascular anatomy of perimedullary AVMs depending on their localisation.

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Table 1: Distribution of Perimedullary Vascular Malformations.

Type		Vascular Structure		Relationship with Spinal Cord	No. of Patients
		Feeders	Draining Vessels		
IPAVF	Cervical IPAVF	Radiculomedullary artery from vertebral artery	Perimedullary veins	Perimedullary dorsal, ventral	9
IPAVF	Thoracic IPAVF	Anterior, posterior spinal or Radiculomedullary arteries	Perimedullary veins	Perimedullary dorsal or ventral	18
IPAVF	Conus Medullaris IPAVF	Radiculomedullary artery that ascending with roots cauda equine	Perimedullary veins, Radicular veins	Perimedullary dorsal or ventral	7
IPAVM		Anterior, Posterior Radiculomedullary artery, Radiculopial artery	Perimedullary veins	Perimedullary dorsal or ventral	8

Table 2: Types of Interventions.

Type		Types of Interventions			No. of Patients
		Endovascular	Microsurgery	Combined	
IPAVF	Cervical IPAVF	2	4	3	9
IPAVF	Thoracic IPAVF	2	16	-	18
IPAVF	Conus medullaris IPAVF	-	7	-	7
IPAVM		1	7	-	8

The vascular anatomy of cervical IPAVF varied. In 8 cases, fistula had a massive blood flow (type C) and was fed by the radiculomedullary arteries originating from vertebral artery. Those feeders were short transitive vessels between the vertebral artery and the draining veins, and were drained by the perimedullary veins. In one case two thin vessels originating from both vertebral arteries at C6 level fed the IPAVF. The fistula emptied in huge perimedullary vein which itself drained cranially and emptied in venous system of the posterior fossa. Hemodynamically, this was type B fistula. The draining vein formed a large venous bag that compressed the spinal cord (Figures 1-6).

Thoracic IPAVF's were diagnosed in 18 patients. In most cases (n = 12), they were fed by the anterior spinal artery (Figures 7-9). Other fistulas were supplied by anterior radiculomedullary (n = 2), posterior spinal (n = 3), anterior and posterior spinal arteries (n = 1). Usually IPAVF's of the thoracic region lead to marked distension of all perimedullary veins of thoracic region. In one case, it was type C and in 17 cases type A and B was demonstrated.

Conus medullaris IPAVF had a specific vascular structure. The fistulae were fed by arteries, which are usually run to the medullar cone together with cauda equine roots. Occasionally, the descending anterior or posterior spinal arteries fed IPAVF's. All of them were type A fistulas. It is important, that these IPAVF's were often missed by angiography. In two cases, fistulas were not revealed during a complete angiography. All conus medullaris malformations were true fistulas. We did not observe any vascular conglomerates or nidus as well as no invasion in conus medullaris tissue. Fistulas looked like a simple contact between the feeding artery and the perimedullary draining veins (Figures 10 and 11).

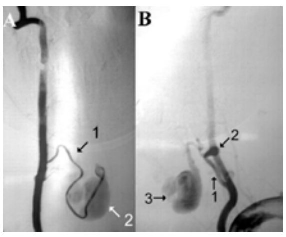

Figure 1: Cervical IPAVF at level C5–6. A. Right vertebral angiogram. B. Left vertebral angiography: 1 – feeding AVF artery; 2 – aneurysm on feeding artery; 3 – initial part of a draining vein.

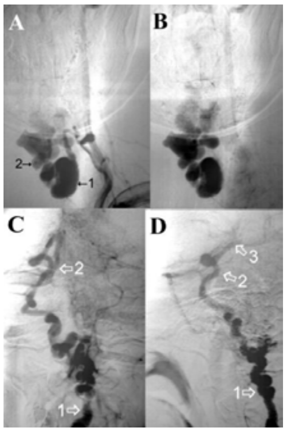

Figure 2: Cervical IPAVF at level C5–6. Left vertebral angiogram. A, B. The initial part draining veins: 1– huge venous pocket, 2 – conglomerate draining veins. C, D drainage to the venous system of the posterior fossa: 1– perimedullary vein, 2– lateral mesencephalic vein, 3– straight sinus.

All our IPAVMs were localized in a thoracic region of the spinal cord. They represented conglomerates of vessels localised intradurally but extramedullary. It looks like nidus with precisely separated from it feeding and draining vessels (Figures 12-16). The following features distinguished IPAVM from IPAVF: 1) IPAVMs always contained conglomerate vessels, but IPAVF's never had true vascular conglomerate, however, occasionally we observed large tortuous draining vessels; 2) in IPAVMs, conglomerate vessels were localized

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near feeding artery, but in IPAVF, the draining vessels were straight forwarded near a fistula, and then they become tortuous more distally (artery empties in a wide vein, which distally becomes more distended and tortuous); 3) IPAVM nidus is drained by perimedullary vein/

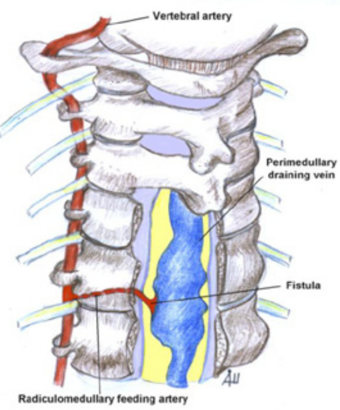

Figure 3: Artist drawing demonstrating vascular structure of cervical IPAVF.

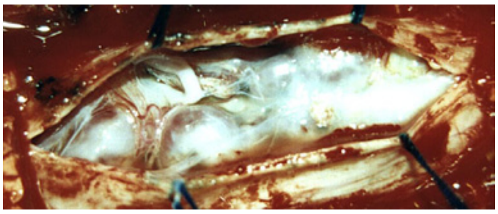

Figure 4: Cervical IPAVF at level C5–6. Intraoperative photo. The huge perimedullary draining vein.

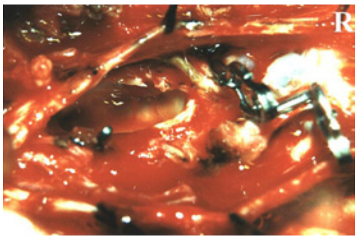

Figure 5: Cervical IPAVF at level C5–6. Intraoperative photo. The perimedullary draining vein opened. Two clips applied cranially because intense retrograde venous bleeding from the venous system of the posterior fossa.

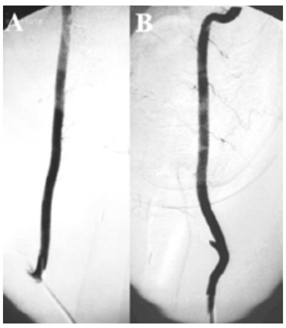

Figure 6: Cervical IPAVF at level C5–6. Angiography after surgery. A right vertebral angiogram. B Left vertebral angiogram.

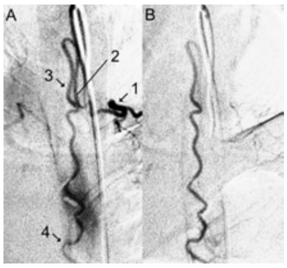

Figure 7: Thoracic IPAVF. Spinal angiography. A. B. Injection in left Th10 segmental artery: 1 – left segmental artery, 2 – anterior radiculomedullary artery, 3 – anterior spinal artery, 4 - anterior spinal artery passes to draining vein.

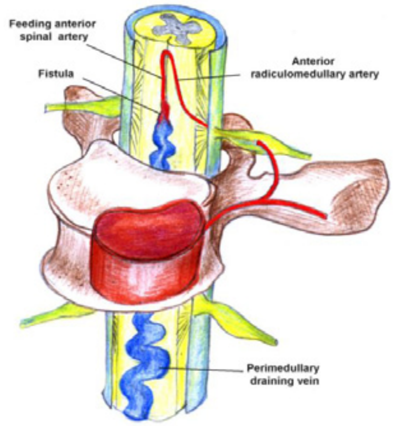

Figure 8: Artist drawing demonstrating vascular structure of thoracic IPAVF.

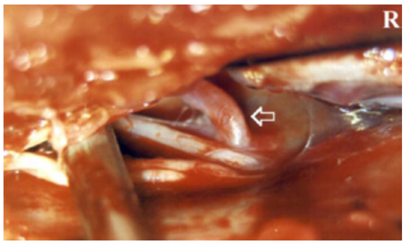

Figure 9: Thoracic IPAVF. Intraoperative photo. The spinal cord rotated, anterior radiculomedullary artery (white arrow) was found among nerve roots: R – rostral (cranial) pole of the wound.

veins which are straight and, therefore, contrasts with a nidus; 4) the number of feeding arteries is larger in IPAVM (n = 3 or more) compared to IPAVF (n = 1-2).

Treatment strategy

In 2 cases cervical IPAVF's, open surgical intervention were dangerous due to high blood flow from vertebral artery to draining vessels. In these cases we performed the endovascular obliteration of the distal portion of feeding vessels by detachable balloons and glue compositions near the place where these vessels empty the draining veins. These interventions allowed keeping the vertebral artery opened. In 4 cases cervical IPAVF's were performed open surgical interventions. In other 3 cases performed combined treatment. In interesting case of cervical IPAVF, which was fed by two vessels

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and drained by huge perimedullary vein with extensive blood flow, initially we carried out an endovascular embolization with glue. However, it was not possible to completely block the feeding vessels. We considered such an occlusion to be inadequate for preventing the recurrence of cervical IPAVF. Thus, a microsurgical intervention was performed as a second phase of the surgical treatment. Large dorsal tortuous veins compressing the spinal cord were detected during open intervention (Figure 4). Two feeding arteries were found and coagulated near the fistula and the venous bag was resected. During this resection an intense retrograde venous bleeding started from the venous system of the posterior fossa. Microsurgical clips were applied on the draining vein at the C2 level and then the venous collector was totally removed (Figure 5).

In 16 thoracic IPAVF's of type A and B, we performed open microsurgical interventions. Endovascular embolisation of enlarged feeding anterior spinal artery with high blood flow (type C) was carried out in two cases. The following microsurgical technique was used. Enlarged perimedullary veins around the spinal cord were found after the opening of the dura. Then, we searched for a place where the feeding artery emptied into the perimedullary veins. Sometimes the tortuous veins complicated the search of the fistula's location. In such cases, the "untangling" of the perimedullary veins technique was applied. The angiographic data was compared with the surgical findings. At first, the feeding radiculomedullary artery was identified. The radiculomedullary artery may feed IPAVF itself or it entered the anterior or posterior spinal arteries and these arteries feed the IPAVF. The radiculomedullary arteries were usually revealed without problems because they followed the spinal roots (Figure 9). Checking the distal part of the artery always helped to find the fistula. When the fistula was fed by an anterior radiculomedullary or anterior spinal artery, we removed the facets joints with a drill unilaterally. Then, sutures placed on the pia at the site dentate attachment and spinal cord was rotated. This approach allows watching and manipulating with anterior radiculomedullary or anterior spinal arteries. The small microsurgical clips were applied to the feeding artery near the fistula. A few minutes after the fistula was isolated and switched off, the perimedullary veins became flat and changed red colour into a cyanotic one. Then, we coagulated and cut the feeding artery. To prevent the reoccurrence, it was essential to cut the distal portion of the feeding vessels at the level where they enter the draining

Figure 10: IPAVF conus medullaris. Intraoperative photo demonstrating the feeding artery ascending with roots of cauda equina and emptying into posterior perimedullary draining vein.

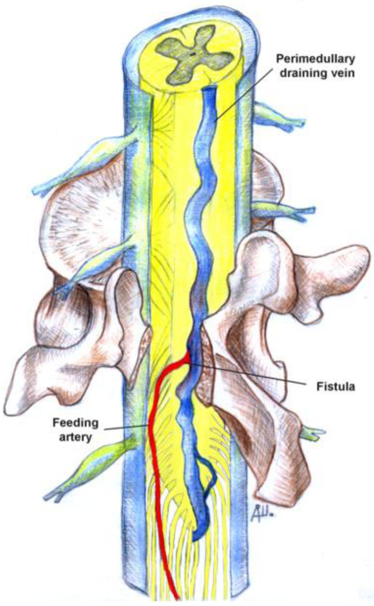

Figure 11: Artist drawing demonstrating vascular structure of conus medullaris IPAVF.

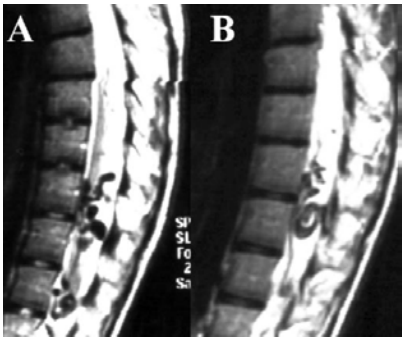

Figure 12: IPAVM at Th10 – L2. T2–weighted MR images. A, B sagittal MR images. A conglomerate of vessels is localized only perimedullary.

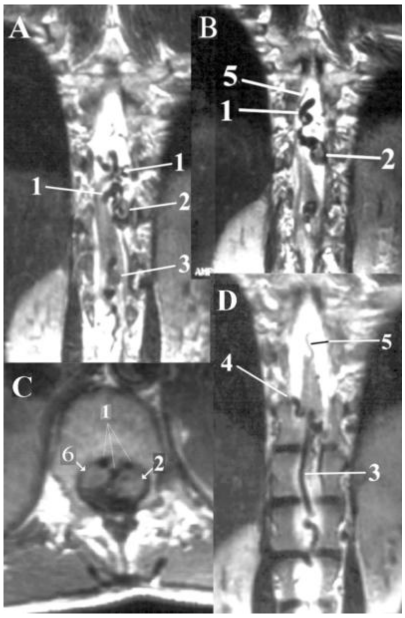

Figure 13: IPAVM at Th10 – L2. A, B, D – T2–weighted sagittal MR images. C – axial T1–weighted MR image: 1 – vessel of a conglomerate; 2 – thrombosed aneurysm; 3 – draining ventral vein; 4 – left L1 feeders; 5 – draining vessel.

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veins. The coagulation was easy at dorsal surface of the spinal cord, although it was very difficult at the ventral surface, where we used the small microsurgical clips. For the type A fistulas, that was the end of surgical intervention. In type B fistulas, enlarged tortuous draining veins caused the spinal cord compression. In 2 cases, these veins were partially removed. The resection of such veins along 1-2 vertebrae did not cause any neurological consequences. After switching of the main fistula, we carried out a search for additional feeding vessels.

The conus medullaris IPAVF's were treated only by open microsurgery (Figure 10). There were not special complexities during intervention on these fistulas. The site of contact of feeding artery with perimedullary veins was coagulated and cut. In ventral conus medullaris IPAVF's, it was easy to perform a rotation of the conus medullaris and switch off the fistulae.

For the treatment of IPAVM we used both endovascular and microsurgical techniques. In one patient, the IPAVM had a large size and high blood flow that required an endovascular intervention. This IPAVM had two feeders. The huge perimedullary conglomerate was supplying from Th9 radiculopial artery and posterior spinal artery, which in turn was supplied by L1 posterior radiculomedullary artery. We reached both feeders by superselective catheterization and embolized with liquid glue (ONYX). In 7 large IPAVMs, we used a microsurgical technique. The posterior approach and the spinal cord rotation were used when necessary. The perimedullary conglomerate of IPAVM usually covered its feeders and draining veins. At the very first phase of surgery, this caused difficulties with revealing the feeders around the conglomerate. Using angiograms for guidance we examined all vessels, which went together with the spinal roots at a level of the malformation and at a level one vertebra above and below. If we found the feeding arteries, we clipped them temporarily.

Recently we have invented a method of "untangling" of the perimedullary conglomerates, which helps to locate the feeding and draining vessels as proximally to vascular conglomerate as possible. We examine these vessels thoroughly to be sure they are not involved in spinal cord blood supply. Thereafter the temporary clips were imposed to feeding and draining vessels as close as possible to a vascular conglomerate. When feeding and draining vessels have branches to a spinal cord, clips were imposed to the artery as distally as possible and at the vein most proximally to the level where the vein

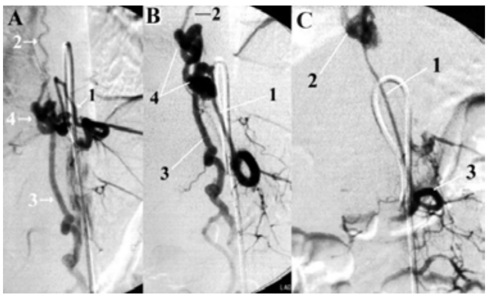

Figure 14: IPAVM at Th10 – L2. Spinal angiography. A. Injection in left Th10 radiculomedullary artery: 1 – feeding left posterior Th10 radiculomedullary artery; 2 – draining vein in cranial direction; 3 – draining vein in caudal direction; 4 – vascular perimedullary conglomerate. B. Injection left Th12 radiculomedullary artery: 1 – feeding posterior Th12 radiculomedullary artery; 2 – cranial draining vein; 3 – caudal draining vein; 4 – vascular conglomerate. C. Injection left L1 radiculomedullary artery: 1 – long ascending feeding L1 posterior radiculomedullary artery; 2 – vascular perimedullary conglomerate; 3 – segmental left L1 artery.

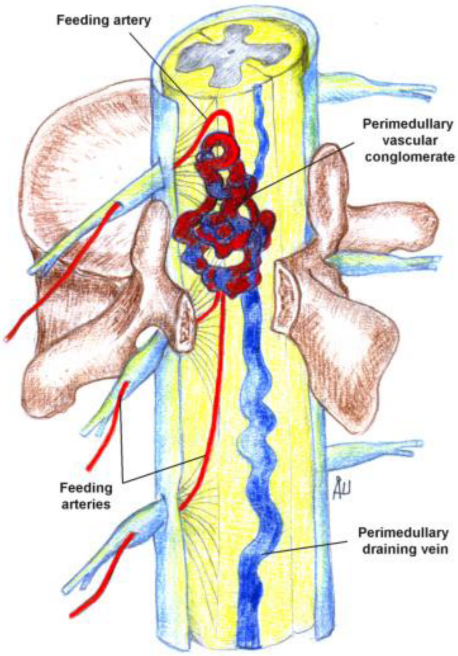

Figure 15: Artist drawing demonstrating vascular structure of IPAVM.

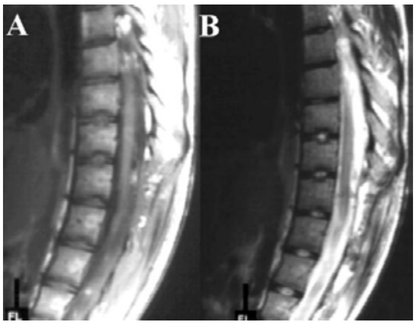

Figure 16: IPAVM at Th10 – L2. MRI after microsurgical resection of the AVM. A T1–weighted, B– T–2 weighted sagittal MR images.

received the branches from a spinal cord. During surgical procedure we separated the feeders and draining vessels at a great distance, and were always able to safely impose clips and thus prevent bleeding. If the clips were applied to the vessels without their proper separation and mobilization, there was a high risk of the rupture with insufficient space for the second application of the clip. The next step was the separating of vascular conglomerate from the spinal cord. During removal of vascular conglomerate we tried to preserve the pial vascular net of the spinal cord when possible. Thereafter the feeders and draining vessels were coagulated, all temporary clips were taken away and the vascular conglomerate was removed (Figure 16).

Treatment results

Postoperative spinal angiography and MRI were used to examine the efficacy of the surgery.

The total obliteration or removal was accomplished in 38 IPAVF's and IPAVMs: 9 cervical IPAVF's, 7 conus medullaris IPAVF's, 16 thoracic IPAVF's and 7 thoracic IPAVMs. In three patients (two thoracic IPAVF and one thoracic IPAVM), the intervention was finished by an incomplete obliteration or removal of malformation.

The clinical results of surgical interventions in relation to localisation and anatomy of IPAVF's and IPAVMs are presented in Table 3. The neurological changes after intervention were assessed using the ASIA scale. In 9 patients, there was a significant regress of neurological signs. They demonstrated increased volume of movements and improved sensitivity. We observed a partial regress of neurological symptoms in 21 patients and no improvement in 9 patients. Three patients had neurological deterioration (Table 3). The most positive results of surgical interventions were obtained in cervical IPAVF's. The worst results were observed in IPAVMs.

Discussion

The treatment strategy in perimedullary vascular malformations depends on their localisation, anatomy and blood flow. The vascular anatomy of the perimedullary vascular malformations was always a field for research [3,4]. Recently Spetzler and his co-workers subclassified them as intradural ventral arteriovenous fistulas. Authors concluded that all such lesions are feeding directly by the anterior spinal artery [4]. However in some cases, we observed perimedullary vascular malformations localised dorsally and those were fed by posterior spinal artery. The perimedullary malformations have long been considered as the fistulae [1]. Recently it was discovered that they might occasionally be real AVMs [5]. In our study (n = 42), 34 patients had IPAVF, and 8 patients had IPAVM. The group of patients with fistulas was also heterogeneous.

In cervical fistulas feeding by vertebral artery, it is possible to successfully obliterate feeding radiculomedullary artery and keep the VA open [4]. Although the best treatment strategy is an endovascular embolisation [6], many of our patients essentially required surgical intervention. There were several indications for surgery including enlarged perimedullary tortuous veins that compressed the spinal cord and retrograde blood flow from venous system of the posterior fossa to perimedullary veins. The goal of microsurgery was to obliterate the fistula and to excise enlarged tortuous draining veins. In all cases it was essential that the distal portion of feeding vessel should be disconnected at the level where it is emptying into the draining veins [3].

Disconnection of the thoracic IPAVF from the spinal venous drainage is currently provided by embolization and microsurgery. The selection of a particular technique depends on blood flow values and on vascular anatomy of fistula. Adequate endovascular treatment requires a number of circumstances to be successful: use of liquid embolic material; penetration of embolic material into the AVF or the proximal draining vein; angiographic disappearance of the AVF after treatment; no compromise of the arterial supply and venous drainage of the spinal cord. Achieving all of these criteria may be difficult. Therefore, in small fistulas with low blood flow, such as type A and B, endovascular intervention is not recommended [7]. In these cases, the surgical intervention would be a method of choice [8]. In fistulas with a high blood flow (type C), the best treatment is endovascular intervention [9,10]. If endovascular treatment does not provide complete obliteration of the fistula, the patient ultimately requires surgery [2].

The conus medullaris AVFs were described earlier. Spetzler and co-workers included them in a modified classification as conus AVM [1]. In our patients, all conus medullaris malformations were fistulas. We did not observe any vascular conglomerates or penetration of malformed vessels in spinal cord. These fistulas were simple contacts of feeding arteries with draining veins.

The IPAVF of spinal cord cone (conus medullaris) could not be approached by endovascular route via cauda equina arteries. Moreover, the majority of such fistulas were those of types A and B. The most effective treatment, therefore, was an open surgical intervention [11,12].

Intradural perimedullary AVMs should be treated only by microsurgical technique, because even after complete devascularization by endovascular technique under the angiographical control, there always remain numerous tiny additional feeders that will eventually enlarge and the blood flow restores. These lesions also cause compression of the spinal cord. However in one of our IPAVMs with high blood flow we had to perform an endovascular intervention as the first step. Later, this patient refused planned microsurgical intervention.

The efficacy of endovascular and microsurgical interventions differs between various types of perimedullary vascular malformations. The highest efficacy was noticed in cases with a cervical IPAVF and conus medullaris IPAVF. The main reason is that their feeding arteries almost never participate in feeding of spinal cord [1,13]. Thoracic IPAVMs and IPAVF's often share blood supply with the

Table 3: Clinical Result of Treatment Intradural Perimedullary Vascular Malformations. The neurological regress was estimated according to ASIA scale. The prominent neurological regress was ascertained if average of impaired motor function in tested muscles improved on 3 - 4 grades, or average of impaired sensation function in tested dermatomes improved on 2 grades. Partial neurological regress was ascertained if average of impaired motor function improved on 1 - 2 grades, or impaired sensation function improved on 1 grade.

Type		Neurological outcome				No. of Patients
		Prominent Improvement	Partial Improvement	No changes	Deterioration	
IPAVF	Cervical IPAVF	3	5	-	1	9
	Thoracic IPAVF	4	8	6	-	18
	Conus Medullaris IPAVF	2	4	1	-	7
IPAVM		-	4	2	2	8
Total		9	21	9	3	42

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spinal cord, and therefore their total resection or obliteration was not always possible [14,15]. Also the most negative clinical outcome was observed in thoracic IPAVF's and IPAVMs [16,17]. In such patients, we usually had no appreciable regress of neurological deficits and there were even few cases with neurological deterioration.

Conclusions

The intradural perimedullary vascular malformations represent a heterogeneous group of arteriovenous malformations with various localisation and vascular anatomy. For surgical purposes it is necessary to distinguish intradural perimedullary arteriovenous malformations (IPAVM) from the intradural perimedullary arteriovenous fistulas (IPAVF). The IPAVF's are subdivided on cervical IPAVF's, thoracic IPAVF's, and conus medullaris IPAVF's. The selection of appropriate surgical strategy depends on the localisation, vascular anatomy, and blood flow rate. In all cases, it is essential to disconnect the distal portion of feeding vessels as much as close to the place where they empty into the draining veins. Whenever possible, the best way is to perform an endovascular occlusion. The microsurgery was reserved for cases when endovascular therapy failed or was unsafe. The highest success rate of obliteration/resection of intradural perimedullary vascular malformations and the best clinical results were observed in the cases of low vertebral IPAVF's and conus medullaris IPAVF's.

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