

A Case of Vestibular and Oculomotor Pathology from Bilateral AICA Watershed Infarcts Treated with Basilar Artery Stenting

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Introduction:

Bilateral AICA infarcts may be the result of impaired arterial flow in watershed territories that overlap with PICA and SCA brainstem/cerebellar circulation among patients with critical basilar artery stenosis (1-3). We report one such patient with watershed bilateral AICA infarcts. She had a two-week history of progressive truncal ataxia, frequent falls, dysarthria and episodic vomiting. Examination suggested brainstem/cerebellar localization. She had bilateral symmetric infarcts of the cerebellar flocculus and middle cerebellar peduncles (MCP) due to tandem proximal and mid-basilar artery (BA) stenosis. Failure to improve on maximal medical therapy led to BA angioplasty/ stenting, with improved brainstem/cerebellum circulation and neurologic deficits.

Case Report:

A 69 year-old hypertensive woman had a two-week history of progressive imbalance, recurrent falls and dysarthria. On examination, she was able to sit without support but required a walker to stand and ambulate. She had normal strength and was without lateralizing signs or limb ataxia. Direction changing horizontal (h) nystagmus without gaze paresis or rebound nystagmus was noted in lateral gaze, and upbeat nystagmus in up gaze. Saccadic h and vertical (v) pursuits with failure to suppress the vestibulo-ocular reflex (VOR) were found. The horizontal head impulse test (h-HIT), positional testing and bithermal caloric responses were normal. Pure tone audiometry showed mild

presbycusis. She had an 8-prism-diopter, comitant esotropia at distance, without skew deviation. The brain MRI showed right and left symmetric cerebellar flocculus and MCP infarcts (Figure 1). Cerebral angiography showed tandem, proximal (85%) and mid (50%) BA stenosis (Figure 2 upper and lower arrows respectively). We found fetal origin of right posterior cerebral artery and absence of supratentorial arterial collaterals. Despite a 12-day course of anticoagulation with heparin, and aspirin, her truncal ataxia worsened. She became unable to sit without support. In addition, episodic, non-positional vomiting developed. BA angioplasty followed by stenting was performed successfully thirteen days after admission. Two wingspan stents, 2.5 mm in the proximal and 3 mm in mid basilar artery were deployed. BA angiography after the procedure showed no residual stenosis.

Six months later, she had a negative Romberg test but was unable to walk in tandem. Horizontal and vertical saccadic pursuits and failure to suppress the VOR remained. The horizontal gaze evoked nystagmus and esotropia resolved. A brisk, sustained, biphasic, initially ageotropic non-paroxysmal, horizontal, right beat-nystagmus developed in the supine position with left head turn; it was not associated with vertigo (Figure 3). Currently, she generally avoids the supine position. She has no other deficits.

Discussion:

Rapidly progressive symmetric vestibulo-cerebellum dysfunction initially suggested in our patient a viral, inflammatory or autoimmune etiology. Imaging however readily identified advanced multifocal BA stenosis as the cause of bilateral symmetric infarcts of the MCP and cerebellar flocculus (Figure 1). We found three previous case reports of such watershed, bilateral AICA strokes (1-3). To our knowledge, we are the first to

improve AICA territory circulation with intra-basilar artery double-stents in this clinical setting.

The horizontal pursuit abnormality and VOR suppression failure found in this case may be seen in isolated lesions of the flocculus in humans (4). We did not find lateral gaze holding weakness; or rebound nystagmus. The slow phase of the horizontal (h) nystagmus did not exponentially decrease in lateral gaze, suggesting a vestibular-nystagmus mechanism. Episodic vomiting in the absence of primary gaze nystagmus in our case may be explained by bilateral simultaneous, symmetric vestibular-cerebellum lesions. Alternatively, area postrema ischemia could be responsible.

Nystagmus in AICA strokes may represent either ischemic peripheral or central vestibular lesions. Direction changing h-nystagmus and a normal h-HIT pointed to central localization (5, 6) implying bilateral sparing of the labyrinth, vestibular nerve and the vestibular root entry zone. We did not perform a perfusion scan; therefore, it is not possible to exclude additional ischemic-penumbra contribution to the clinical findings. Most of the findings observed, however, may be attributed to lesions involving bilateral cerebellar flocculus and cerebellar projections within the MCP. A mild esotropia may be found in patients with cerebellar lesions due to vergence dysfunction.

Our patient had also sparing of the inner ear circulation (internal auditory arteries), usually an AICA branch. Previously reported large AICA infarct series have shown a consistent clinical manifestation of mixed peripheral and central vestibular abnormalities and frequent cochlear dysfunction. Severe cochlear loss has been reported previously with watershed AICA infarcts (3).

The vascular anatomy and mechanism of infarction in this case are interesting and different from previously reported AICA watershed infarcts (1-3). Whereas the clinical manifestations of BA stenosis /occlusion may be occasionally asymptomatic, a rapidly evolving neurologic deficit is common. Presentations over several days/weeks are less frequent (10). Large arterial occlusion, dissection artery to artery emboli is the common stroke mechanisms in vertebro-basilar artery disease. Subacute, bilateral watershed AICA infarcts due to tight tandem BA stenosis are unusual (1-3). The patency of PICA, AICA and SCA was demonstrated in our patient prior to and following BA stenting. Fetal origin of the right posterior cerebral artery and lack of substantial collaterals from the anterior circulation undoubtedly contributed to the pathogenesis of the stroke. We did not observe positional changes in the neurologic examination although she preferred to stay in the supine position. Clinical worsening despite maximal medical therapy led to BA angioplasty/ stenting

The histologic characteristics of the basilar artery make the combination angioplasty /stent the best intravascular intervention choice. An insubstantial BA muscularis and adventitia layers, limits the intraluminal pressure range to be exerted during angioplasty, due to a high risk of perforation (). In addition, the stent provides support to the vessel wall and does not seem to occlude perforator vessels. Accordingly most reported case series in the past decade utilized combined angioplasty with stent placement.

Presently, the level of evidence is lacking when considering routine intra-basilar angioplasty/stenting. However, there is an increasing body of literature suggesting angioplasty-stent placement as a generally acceptable option, given very grave prognosis in patients with rapidly deteriorating neurologic status from advanced BA stenosis.

Moreover, following the first reported successful placement of coronary artery stent in the BA in a patient with on-going deterioration despite anticoagulation (), four reported case series of symptomatic advanced BA stenosis, suggest a significant rate of success, however, the frequency and severity of major complications needs further assessment (table 1) (7-11). Currently, a prospective, randomized clinical trial designed to compare maximum medical therapy versus maximum medical therapy with stenting begun in 2008. This on-going study will conclude once 180 patients are enrolled. It is currently in progress (12). Therefore, until more data is available, intra-arterial interventions such as stenting should only be considered on the basis of perhaps the last resort among conventional maximal, medical therapy failures. The patient's clinical temporal profile may define the indication. We wish to add this case to the literature not only to illustrate the unusual presentation and the pathophysiology of infarction, but also to report a favorable outcome with interventional treatment.

FIGURE 1 Axial Diffusion weighted DWI- MRI of the brain, 3 mm cuts. Image A shows areas of increased signal involving the cerebellar flocculus bilaterally. Image B shows increased signal involving the lower section of the middle cerebellar peduncles bilaterally. Image C shows bilateral increased signal in the upper section of the middle cerebellar peduncles bilaterally. The infarcted tissue in sections A and B are symmetric; In the upper C section left infarct is greater than right. The ADC map (not shown) confirmed restricted diffusion. The infarcted territory involves the watershed between PICA, AICA and superior cerebellar arteries

FIGURE 2 Pre and post Stent Cerebral Angiography. Panel A shows tight stenosis of the proximal (lower arrow) and less severe mid-basilar artery (upper arrow) stenosis, proximal to the origin of AICA arteries. Panel B post-insertion of wing stent shows successful re-canalization of the basilar artery

Figure 3

VNG obtained in darkness with the patient in the supine position with the head turned to the left. A right-bat nystagmus developed for ~ 10 sec and changed direction a non-rhythmic fashion as long as the head maintained this position. The patient was asymptomatic.

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