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Clinical and neuroradiological characteristics of ischemic stroke and subarachnoid hemorrhage in isolated posterior inferior cerebellar artery dissection: Literature review and report of 2 cases

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ABSTRACT

Background/objective: Isolated dissection of the posterior cerebellar artery (PICA) is very rare, with only few cases reported in the literature. Ischemic stroke or subarachnoid hemorrhage (SAH) may follow isolated PICA dissection depending on the dissection plane of the artery. The aim of this is study is to investigate similarities and variations in clinical, neuroradiological manifestations as well as the management and prognosis of ischemic stroke and SAH events in isolated PICA dissection.

Method: We report 2 cases of PICA dissection treated at our institution, one with ischemic stroke and other with SAH. Peer reviewed articles were used for insight on differences and similarities in presentation, management and prognosis between PICAD with ischemic stroke and SAH event.

Results: Occipital headache was noted to be the most common clinical presentation in both patients groups. Patients with SAH event presented with more severe symptoms compared to those with ischemic stroke. Susceptibility weighted imaging (SWI) was found to be better at detecting PICAD compared to conventional MRI/MRA. Digital Subtraction Angiography remains the gold standard diagnostic test for dissection. PICAD with ischemic stroke is likely under diagnosed.

Conclusion: PICAD with ischemic stroke is likely under-diagnosed due to the following reasons; 1) patients present with neurological deficits that are non-specific and mild, 2) DSA, which is a gold standard of diagnosing PICAD is not regularly performed in these patients, 3) lack of predisposing factors associated with PICAD in some patients. The mild and non-specific clinical presentation in young adults with PICAD with ischemia probably due to their ability to compensate better given low incidences of other comorbidities. Further investigation regarding the underlying etiology of isolated PICAD, association of proximal PICAD to ischemia and distal PICAD to SAH as well as use of SWI in detecting PICAD is recommended.

1. Introduction

Isolated dissection of the posterior inferior cerebellar artery (PICA) is a relatively uncommon event. Given the small diameter of this artery, it is difficult to diagnose a PICA dissection (PICAD). Few cases of isolated PICAD are reported in the literature [2,4–5]. PICAD is usually associated with increased risk of ischemic stroke and subarachnoid hemorrhage (SAH) [2–3]. Proximal dissections are usually associated with ischemic stroke while distal dissections with SAH [2–5]. Matsumoto et al. [3] attributes the association of distal dissection with SAH to the fact that distal dissections in PICA normally have high frequency of aneurysmal dilation incidences leading to SAH. Per Kaboyashi et al. [2], ischemia secondary to the distal PICA dissection are usually tiny and mild that they can only be detected after aneurysmal changes that regularly lead to SAH. More dedicated studies are however needed to shade more light on the association of distal dissection of PICA with SAH.

Although isolated PICAD present with either SAH or ischemic stroke, there are currently no studies showing the variations in their clinical and radiological presentation and management. In this study, we report 2 representative cases of PICAD that presented with ischemic stroke and SAH. We reviewed literature to show the differences in their clinical and radiological presentation and management.

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2. Review of the literature

Potential eligible reports and studies written in English were identified using PubMed. Terms 'Isolated PICA dissection or PICA and dissection' were used. We reviewed reports/studies and selected patient with ischemic or SAH events following isolated spontaneous PICAD.

3. Case presentation

3.1. Case #1

A 52-year-old female presented with a 1-day history of nausea, vomiting, vertigo, gait instability, and headaches. Her past medical history included hypertension, 3 times cesarean section and septorhinoplasty. On physical examination, she had mild dysmetria, worse on right. Head Computed Tomography (CT) scan and Magnetic Resonance Imaging (MRI) showed an extensive left cerebellar hemisphere infarct (Fig. 1A). CT angiography (CTA) showed focal stenosis in the left PICA, with no flow beyond the stenosis, a beaded appearance of the left vertebral artery and bilateral internal carotid arteries, suggestive of fibromuscular dysplasia (Fig. 1B). Patient was found to have left PICA dissection on CTA, she was started on aspirin, statin, and antihypertensive medication for secondary stroke prophylaxis. She achieved full clinical recovery within 3 months. Her CTA after 7, 12 and 18 months showed persistence of the beaded appearance of the left PICA with improvement and recovery of flow along the distal PICA (Fig. 1C).

3.2. Case #2

A 60-year-old female presented with a 3-day history of headaches, nausea, vomiting, loss of consciousness, seizure, and a Glasgow Coma Score (GCS) of 9. On head CT, she had basal SAH with intraventricular hemorrhage (IVH) and hydrocephalus (Fig. 1D). CTA and Digital Subtracted Angiography (DSA) showed a 2 × 3.5-mm dissecting aneurysm along the proximal left PICA (Fig. 1E). The patient underwent External Ventricular Drain (EVD) placement to relieve the hydrocephalus and IVH, followed by permanent ventriculo-peritoneal shunt placement due to multiple unsuccessful attempts of EVD weaning. 9 months after the initial event, she underwent endovascular treatment of the aneurysm through placement of a 3.25 × 14 mm Pipeline embolization device (PED, Pipeline Flex, Medtronic, MN, USA) in the vertebral artery segment where the PICA originated. 3 months later, a second 3.25 × 14 mm PED was placed in the left vertebral artery overlapping PICA origin to improve aneurysm occlusion. The interventions had no complications. Follow-up CTA 1 year after the last procedure showed patent PED stent, normal flow within the PICA, and minimal decrease in size of PICA aneurysm (Fig. 1F), accompanied by full recovery except for mild cognitive impairment.

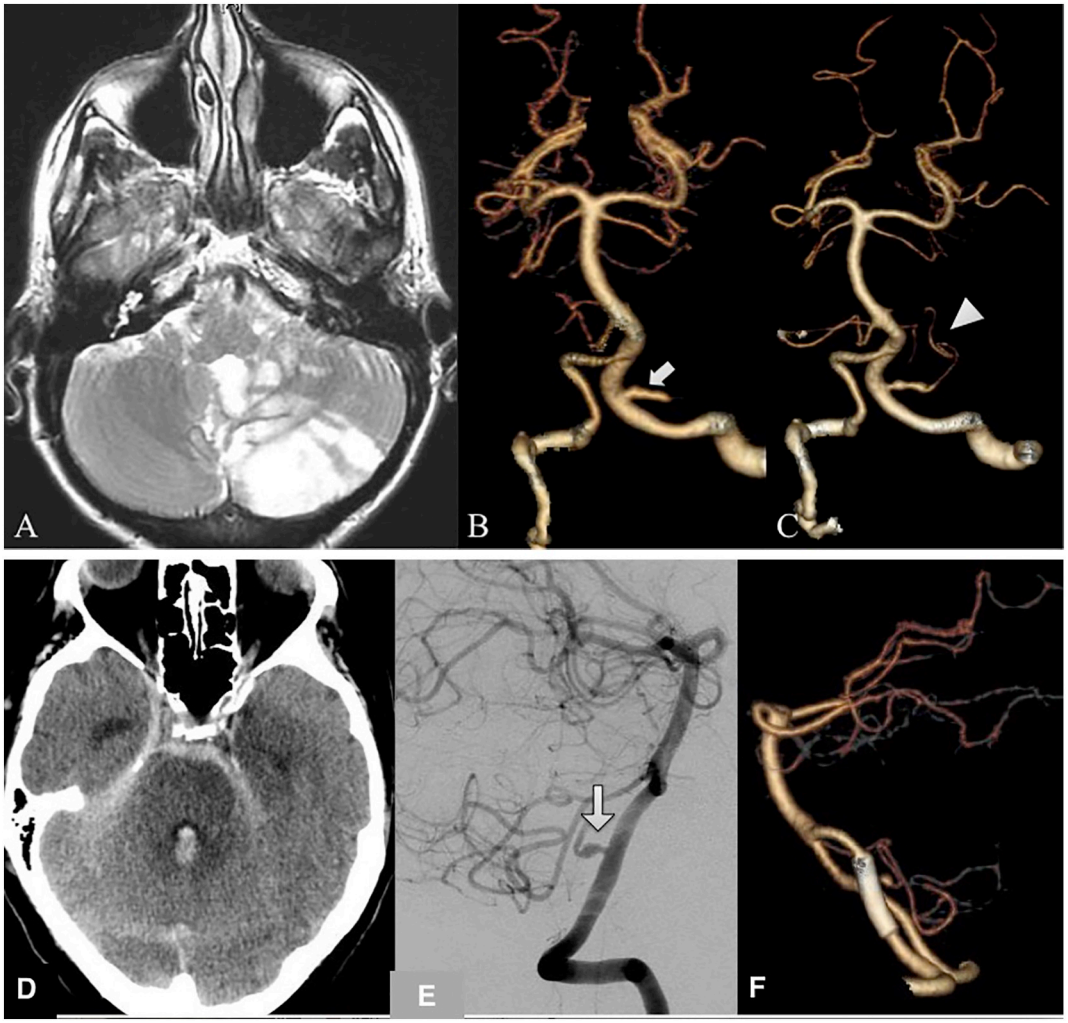

Fig. 1. Imaging of case 1. MRI (A) and CTA (B) at presentation show cerebellar infarct due to left PICA dissection (arrow). CTA 1 year after conservative treatment (C) shows recovery of flow (arrowhead). Imaging of case 2. CT (D) and CTA (E) show SAH due to a dissecting aneurysm in left PICA (arrow), which was treated by pipeline stent placement (F).

4. Discussion

Despite an increase in diagnosis of isolated PICAD in the recent years due to advances in imaging studies, it remains a rare vascular pathology [2–5]. The underlying cause of isolated PICAD is not well understood, etiologies such as hypertension, trauma, connective tissue disorders and specific arteriopathies have been suggested as risk factors. However, there are mixed reports in the current literature regarding association of the above suggested risk factors with isolated PICAD, with some authors reporting lack of association of the suggested risk factors with isolated PICAD. [2–6]. It is important to mention that to our knowledge there have been no systematic studies performed to understand the underlying causes of isolated PICAD, including the ones quoted here. In one of our patients, beaded appearance of the vertebral artery and carotid arteries was suggestive of fibromuscular dysplasia and both patients had a past medical history of hypertension.

Majority of PICAD occur on the left side, probably due to the tendency of the left vertebral artery dominance [2–5]. Proximal segment of PICA is the most frequently dissected portion and is associated with ischemic stroke while distal dissection is associated with SAH [2–5]. However, higher percentages of cases of PICAD with SAH event have been reported in literature [3], raising the question of whether PICAD with ischemic stroke is being under diagnosed. Of note, DSA; the gold standard for diagnosing PICAD, is not routinely performed in ischemic stroke [2–5].

In literature review, patients with PICAD present between the ages of 22–68 years; the median age of onset is 43.8 years. Men and women seem to be affected equally. All patients (n = 35) presented with occipital headache as first presenting symptom followed by symptoms involving the medulla or cerebellum. All SAH patients presented with severe headache sometimes with LOC while Ischemic stroke group presented with neurological deficits that were generally non-specific and mild, hence the suspicion that maybe PICAD associated with ischemia under diagnosed. Since DSA, which is, a gold standard for diagnosis of PICAD is not regularly performed in these patients. Our two patients presented with generalized headaches not occipital headache. Additionally, unlike the patient with PICAD associated SAH, the patient with PCAD associated with ischemia did not presented with more severe symptoms such as seize and LOCs. She also had a better prognosis attaining full clinical recovery within 3 months.

5. Imaging

DSA is the ‘gold standard’ imaging modality for the diagnosis of dissection. [2–5]. ‘Pearl and string’ aspect is the most frequent sign, which appears as a smooth segmental tapered stenosis followed by a fusiform or saccular dilatation of the artery [3–4]. Since DSA is not readily performed in ischemic stroke, diagnosing arterial dissection can be challenging. Other imaging modalities used in diagnosing PICAD, include MRI/MRA and CTA. Conventional MRI or fat-suppressed T1-weighted imaging may not easily detect intramural hematoma of PICA because of its small diameter and tortuosity. The same limitations apply to MRA especially with subtle changes; hence, initial MRA alone is not enough to diagnose PICAD. Serial changes of MRA are useful in monitoring changes in PICAD. Some studies found susceptibility weighted imaging (SWI) more helpful in detecting abnormalities of PICA than conventional MRI/MRA due to its ability to detect small vessel abnormalities [3–4]. Subsequent angiography is recommended in dissections with ischemic stroke for pseudoaneurysm evaluation and treatment to prevent hemorrhagic conversion. [2,4–5].

6. Treatment

In PICAD with ischemic stroke, early anticoagulation with heparin or low molecular weight heparin (LMWH) have been advocated at the time of diagnosis [1], since the risk of stroke is greatest in the first few days following initial vascular injury. Antithrombotic drugs, especially anticoagulants, are avoided in the presence of dissecting aneurysms. Based on observation data, antiplatelet therapy is considered safer than anticoagulation [1–5].

In the literature reviewed, all patients with PICAD with ischemic stroke were treated with anticoagulant/antiplatelets. SAH and IVH should be ruled out with imaging before starting treatment [1–4]. Our patient was started on ASA 81 mg daily with full recovery. Clinical course of ischemic type of spontaneous isolated PICAD is relatively benign without significant squeal.

Management of PICAD associated with SAH event includes conservative treatment, endovascular or surgical approach. While there is no standardized way of managing these patients, conservative treatment was generally used for patients with very poor clinical conditions. Most studies report considering embolization as a first treatment option [1–4]. Our patient was treated by successful placement of a pipeline stent in the vertebral artery across the origin of PICA where the aneurysm originated since the aneurysm was located in a perforator rich segment and bypass followed by trapping could result in perforator territory ischemia.

7. Conclusion

PICAD with ischemic stroke is likely under-diagnosed due to the following reasons; 1) patients present with neurological deficits that are non-specific and mild, 2) DSA, which is a gold standard of diagnosing PICAD is not regularly performed in these patients, 3) lack of predisposing factors associated with PICAD in some patients. The mild and non-specific clinical presentation in young adults with PICAD with ischemia probably due to their ability to compensate better given low incidences of other comorbidities along with few vascular risk factors in this cohort compared to older patients puts them even at a higher risk of being under-diagnosed [2]. In spite of DSA being the gold standard for diagnosing PICAD, we recommend that it should be performed at the discretion of the clinicians tailored to individual patient needs while weighing carefully their medical risks and benefits. Given the limited available information regarding isolated PICAD in general, dedicated investigations regarding the predisposing risk factors of PICAD, association of proximal PICAD to ischemia and distal PICAD to SAH as well as use of SWI in detecting PICAD is recommended to better help with prevention and management of PICAD.

Declaration of Competing Interest

No conflict of interest.

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