Marmara Medical Journal 2016; 29: 185-188
DOI: 10.5472/MMJcr.2903.03

CASE REPORT / OLGU SUNUMU

Persistent primitive trigeminal artery as a cause of trigeminal neuralgia and persistent primitive trigeminal artery associated with pituitary adenoma: Two case reports

Trigeminal nevraljiye neden olan persistan primitif trigeminal arter ve pituiter adenoma eşlik eden persistan primitif trigeminal arter: iki olgu sunumu

Furkan UFUK, Duygu HEREK

ABSTRACT

Persistent primitive trigeminal artery (PPTA) is an abnormal embryonic vascular structure that provides connection between the carotid artery and vertebrobasilar system. PPTA usually originates from cavernous segment of internal carotid artery (ICA) and contacts with the central part of the basilar artery. The sellar lesions such as pituitary adenoma accompanying PPTA are very rare and PPTA is an unusual cause of trigeminal neuralgia. Herein, we present two cases of PPTA along with a prolactin secreting pituitary adenoma and trigeminal neuralgia (TN) due to PPTA, diagnosed by magnetic resonance imaging (MRI). We suggest that MRI would be able to properly plan and execute surgical treatment and avoid cerebral digital subtraction angiography (DSA).

Keywords: Magnetic resonance imaging, Persistent trigeminal artery, Prolactinoma, Trigeminal Neuralgia

ÖZ

Persistan primitif trigeminal arter (PPTA), anormal embriyonik vasküler yapı olup karotis arter ve vertebrobasiler sistem arasında bulunur. PPTA sıklıkla internal karotid arterin kavernöz segmentinden kaynaklanmakta olup basiller arterin santral kesimiyle birleşir. Hipofiz adenomları gibi sellar lezyonlara eşlik eden PPTA nadiren görülür ve PPTA, trigeminal nevraljinin nadir bir nedeni olabilir. Biz burada prolaktin salgılayan hipofiz adenomuna eşlik eden PPTA ve trigeminal nevraljiye neden olan PPTA tanısını manyetik rezonans görüntüleme (MRG) ile koyduğumuz iki olgu sunuyoruz. Cerrahi tedavinin planlanmasında ve yol göstermesinde invaziv yöntem olan serebral dijital substraksiyon anjiyografi (DSA)'den kaçarak, yerine MRG'nin kullanılabileceğini düşünüyoruz.

Anahtar kelimeler: Manyetik rezonans görüntüleme, Persistan trigeminal arter, Prolaktinoma, Trigeminal Nevralji

Introduction

Persistent primitive trigeminal artery (PPTA) is one of the abnormal vascular residues between carotid and vertebrobasilar system which is rarely seen in adults. It is detected incidentally during angiography with a prevalence of 0.58 %. PPTA usually originates from cavernous segment of internal carotid artery (ICA) and contacts with the central part of the basilar artery by coursing inside (medial, intrasellar type) or outside (lateral type) the sella [1]. Although patients are usually asymptomatic, it has been reported that it may accompany third and sixth cranial nerve palsies, aneurysms, vascular anomalies and hemifacial spasm [2-4]. The pituitary lesions accompanying PPTA and trigeminal neuralgia (TN) associated with a PPTA are rarely seen [5-10]. The recognition of the presence

Furkan Ufuk (✉)
Radiology Clinic, Sandikli State Hospital, Afyonkarahisar, Turkey
e-mail:furkan.ufuk@hotmail.com

Duygu Herek
Department of Radiology, School of Medicine, Pamukkale University, Denizli, Turkey

Submitted/Gönderme: 15.05.2016 **Accepted/Kabul:** 03.07.2016

of intrasellar PPTA is very important in cases which will undergo transsphenoidal pituitary surgery for pituitary lesions because it can cause life-threatening major bleeding. Knowledge of the typical imaging findings of PPTA prevents major surgical complications [8]. In patients with trigeminal neuralgia lateral type of PPTA should be considered as a cause because these patients can be successfully treated by decompression surgery or embolization of PPTA.

Herein, we present two cases of PPTA along with a prolactin secreting pituitary adenoma and trigeminal neuralgia (TN) due to PPTA and want to emphasize the importance of magnetic resonance imaging (MRI) findings.

Case Report - 1

A 16-year-old female was admitted to the hospital with a 4-month history of headache, galactorrhea and menstrual irregularity. On physical examination, she had normal blood pressure of 120/80 mmHg, pulse of 79 and oxygen saturation level of 99% on room air. There is no history of surgery or drug use. Laboratory investigations revealed an elevated prolactin (PRL) (159.2 ng/mL, normal: 2.1-18.1 ng/mL) level. Blood level of beta human chorionic gonadotropin (β-hcg) was normal. Thyroid function tests showed a normal free thyroxie (FT4) (1,14 ngl/dL; normal 0,78–1,81), free triiodothyronine (FT3) (2,88 pg/mL; normal 2,2–4), serum thyroid stimulating hormone (TSH) (4,28 μIU/mL; normal 0.32–5.4) and serum thyroglobulin antibody (TgAb) (<20,0 IU/ ml; normal 0-40). Complete blood count was normal. Her basic metabolic panel was unremarkable. History of menstrual irregularity, galactorrhea and elevated PRL levels, suggest prolactin secreting pituitary tumor and dynamic contrast enhanced MRI was obtained using a 1.5 Tesla scanner. MRI showed a well-defined ovoid lesion located in the middle part of pituitary gland measuring 6 x 3 mm. The lesion was hypointense on T1 and T2 weighted images and showed a slight enhancement after intravenous contrast media application (Figure 1). A tubular structure extending between cavernous segment of left ICA and middle part of basilar artery by passing through the pituitary gland was observed near the lesion. The appearance suggested PPTA and magnetic resonance angiography (MRA) with maximum-intensity-projection (MIP) images was obtained without any contrast media. MRA (3 dimensional time of flight (3D TOF) MRA) confirmed left intra-sellar PPTA and hypoplasia of basilar artery (Figure 2). The pituitary lesion was considered as microprolactinoma because of its size, typical localization and signal. The patient was treated with a dopamine agonist (bromocriptine) and prolactin, value decreased to normal after 3 months of follow-up.

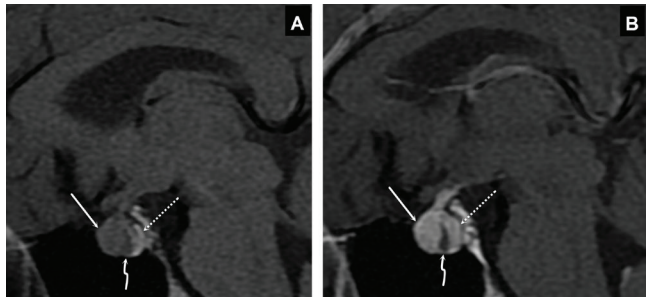

Figure 1: A) Sagittal T1-weighted and **B)** post-gadolinium T1-weighted pituitary MRI scans show pars neuralis (white dashed arrows), pars distalis (white arrows) and hypointense microprolactinoma (white curved arrow).

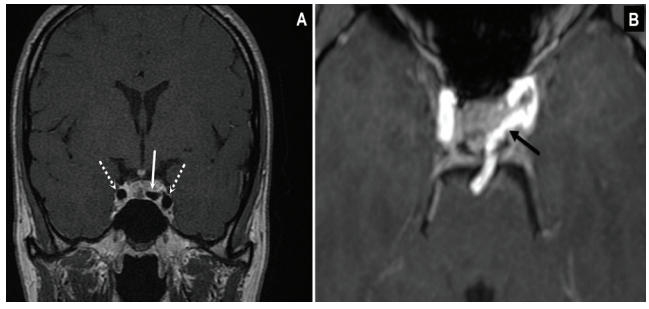

Figure 2: A) Coronal post-gadolinium T1-weighted pituitary MRI scan shows internal carotid arteries (white dashed arrows) and intra-sellar persistent primitive trigeminal artery (white arrow). **B)** Axial post-gadolinium T1-weighted MRI scan shows intra-sellar persistent primitive trigeminal artery (black arrow).

Case Report – 2

A 72-year-old male presented with a six-year history of left facial pain felt in the area supplied by maxillary and mandibular branches of the trigeminal nerve. Pain was described as stabbing, chronic with intermittent flares-up and was not associated with a seasonal pattern. Stimulation of trigger zones by chewing, light touching or shaving, caused the onset of pain. Nonsteroidal anti-inflammatory drugs and carbamazepine were effective for pain relief. However, it became refractory to medical treatment during the past two years. Laboratory investigation was normal. Contrast enhanced MRI was obtained using a 1.5 Tesla scanner. MRI showed a tubular structure extending between cavernous segment of left ICA and middle part of basilar artery by passing through the Meckel's cave. The appearance suggested PPTA and MRA was obtained. MRA (3D TOFMRA) with maximum intensity projection (MIP) images confirmed left lateral type PPTA in contact

with the left trigeminal nerve (Figure 3). Surgical treatment was recommended. After a successful microvascular decompression surgery the patient's pain disappeared completely and had no complaint at 5-month follow-up.

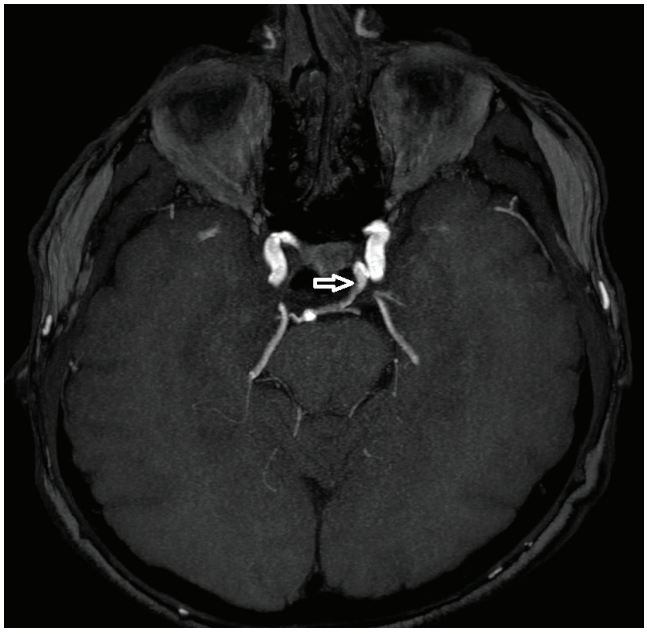

Figure 3: Axial MR-angiography scan shows lateral type of persistent primitive trigeminal artery (white arrow).

Discussion

Trigeminal artery which extends parallel to trigeminal nerve in intrauterine life, supplies the basilar artery because posterior communicating system and vertebral arteries are not fully developed. Normally this artery regresses in the prenatal period, if no regression occurs it is named a PPTA. It usually originates from the cavernous segment of ICA, travels lateral to the sella turcica (lateral type) and reaches the basilar artery or penetrates the sella turcica (medial type) [1]. In our cases, the PPTAs were connected with the left ICA. We observed PPTA penetrating the sella turcica (medial type) in the first case and PPTA traveling to left postero-laterally and inferiorly around the dorsum sella (lateral type) in the second case.

PPTA is usually reported to be detected incidentally and is shown to be rarely associated with pituitary lesions, vascular anomalies, trigeminal neuralgia and aneurysms [3, 4]. Four cases of functional macro-adenoma and one case of non-functional adenoma accompanying PPTA have been reported in the literature [6-9]. PPTA and accompanying prolactinoma is an extremely rare condition and previously only one case has been reported in the English literature [10].

There are several reports about the use of cerebral digital subtraction angiography (DSA) for verification of PPTA in cases of TN due to PPTA or PPTA accompanying pituitary lesions [4-6]. In our cases, however, we used only MRA. Assessment with MRA without doing a cerebral DSA, guided clinicians quite successfully to decide upon surgical treatment. Cerebral DSA has the disadvantages such as radiation exposure, vascular injury and contrast induced nephropathy. MRI should be performed in patients who had TN or with a history suspicious for pituitary lesions. MRI showed PPTA and micro-prolactinoma in our first case, TN due to PPTA in the second case.

Szeifert et al., presented a case of PPTA accompanying a macro-adenoma that caused severe bleeding during operation and emphasized the importance of imaging patients before operation to prevent severe injuries [8]. Abe et al. [9] have detected PPTA during surgery in a case which developed tumoral apoplexy because of non-functional macro-adenoma and they also stated the necessity of preoperative imaging [9]. PPTAconcurrent with hypoplasia of basilar artery is rarely seen in adults with a prevalence of 0.38 % [1]. Careful review of preoperative MRI is important in order to identify pituitary lesions along with uncommon vascular structure. Diagnosis of prolactin secreting adenoma was confirmed by the clinical, laboratory and imaging findings in our first case whereas diagnosis of PPTA was proved by unenhanced MRA.

We suggest that, patients who are candidates for carotid artery surgery or endovascular treatment should be investigated for the presence of PPTA. Because during carotid endarterectomy, a clamp put on PPTA of the same side can cause significant reduction in cerebellar perfusion as well as the brainstem. Emboli originating from carotid artery of the same side or therapeutic endovascular embolic material at the beginning of PPTA may result in ischemia or infarction in posterior circulation. Endovascular treatment is the recommended method in symptomatic cases with PPTA. Endovascular treatment has replaced open surgery to close the abnormal connections. Transarterial or transvenous administration of treatment is done by using balloons, coils and liquid adhesive agents [2].

Conclusion

Unrecognized vascular anomalies such as PPTA in or near the sella turcica lead to severe complications during surgery. When we see a well-defined, round shaped and continuous lesion within or near the sella, PPTA and other

vascular malformations should be kept in mind. When we suspect PPTA, MRA is an applicable method to confirm the diagnosis and prevent surgical complications. If the imaging characteristics are not known well or not enough attention is paid during evaluation, a vascular structure within the pituitary gland may mistakenly be considered as a lesion originating from the gland and this can result in life-threatening complications. Therefore, it is very important to know imaging findings of the vascular anomalies such as PPTA and keep them in mind in the differential diagnosis of pituitary lesions. MRI is useful to confirm the diagnosis and facilitate the surgery in patients with TN. To identify these anomalous arteries on MRA, MIP images are useful. MRA is helpful in proper planning and execution of surgical treatment. Using MRA for diagnosis would allow invasive cerebral DSA to be avoided in patients with suspicious clinical presentation.

References

1. Kalmykov MIu, Ternovoĭ SK. The persistent trigeminal artery as evidenced by multislice spiral computed tomographic angiography. Vestn Rentgenol Radiol 2014;5:19-25.
2. Chen WH, Tsai TH, Shen SC, Shen CC, Tsuei YS. A Case of giant thrombosed persistent primitive trigeminal artery aneurysm presenting with trigeminal neuralgia and successfully treated by a covered stent:Case report and review of literature. Clin Neuroradiol 2015;25:207-10. doi: 10.1007/s00062-014-0314-6.
3. Conforti R, Parlato RS, De Paulis D, et al. Trigeminal neuralgia and persistent trigeminal artery. Neurol Sci 2012;33:1455-8. doi: 10.1007/s10072-012-0942-z.
4. Nishio A, Nishijima Y, Komiyama M, Hara M. Primitive trigeminal artery variant aneurysm treated with Guglielmi detachable coils--case report. Neurol Med Chir 2001;41:446-9. doi.org/10.2176/nmc.41.446
5. Kabatas S, Albayrak SB, Cansever T, Hepgul KT. Microvascular decompression as a surgical management for trigeminal neuralgia: a critical review of the literature. Neurol India 2009; 57:134-8. doi: 10.4103/0028-3886.51279.
6. Lee KS, Kelly DL Jr. Intrasellar persistent trigeminal artery associated with a pituitary adenoma. Case report. J Neurosurg 1989;70:271-3. doi: 10.3171/jns.1989.70.2.0271.
7. Navas M, Martinez P, Shakur SF, et al. Intrasellar chordoma associated with a primitive persistent trigeminal artery. Turk Neurosurg 2015;25:146-53. doi: 10.5137/1019-5149.JTN.8759-13.1.
8. Szeifert G, Lányi F, Czirják S, Pásztor E. Persistent trigeminal artery associated with pituitary adenoma. Orv Hetil 1991;132:1433-4.
9. Abe T, Fujita S, Ozawa H, et al. Haemorrhagic nonsecreting pituitary adenoma associated with persistent primitive trigeminal artery. Acta Neurochir 2000;142:1423-4. doi: 10.1007/s007010070015.
10. Machado MC, Kodaira S, Musolino NR. Persistence of intrasellar trigeminal artery and simultaneous pituitary adenoma: description of two cases and their importance for the differential diagnosis of sellar lesions. Arq Bras Endocrinol Metabol 2014;58:661-5. doi: 10.1590/0004-2730000003363.