

Ruptured intracranial internal carotid artery aneurysm causing subarachnoid hemorrhage and ophthalmoplegia associated with metastatic carcinoma with unknown primary in sellar-parasellar region: True or Coincidental Association

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Abstract: Unruptured intracranial aneurysm can be picked up incidentally on cranial angiography screening for other cause. However, with increasing use of neuroimaging led to increasing rate of pickup and many such incidentally detected cases are treated with endovascular treatment, or craniotomy and clipping of aneurysm or uncommonly a very small aneurysm unsuitable for retreatment by currently available modality may be observed as wait and watch policy. Extremely uncommonly a patient undergoing investigation for suspected subarachnoid hemorrhage following aneurismal rupture, computed tomography scan revealed presence of subarachnoid hemorrhage associated with sellar mass. Such association was previously unreported. In extensive Pubmed search by authors could find a case reported by Yang et al., a case of known surgically resected gastric carcinoma developing secondary in sella, CT angiography of the intracranial artery revealed an aneurysm of ICA located at the inner edge of the siphon segment. Authors report an interesting case of 65 - year -old male presented with aneurysmal subarachnoid hemorrhage with secondary in sella with unknown primary, CT Scan showed presence of sellar mass, underwent digital subtraction angiography at our centre, revealed presence of right supraclinoid internal carotid bifurcation aneurysm, underwent craniotomy and successful surgical clipping of aneurysm with gross total decompression of sellar and suprasellar mass To the best knowledge of authors, current case represents first case in western literature, ,who presenting first time

with aneurysmal subarachnoid hemorrhage and coincidentally associated with incidental sellar mass of metastatic origin Pertinent literature is discussed briefly.

Key words: Adenocarcinoma, aneurysm, secondary in sella surgical management, primary lesion, association

Introduction

Intracranial aneurysm can be detected after rupture or rarely picked up incidentally. The ruptured intracranial aneurysm present with sudden severe headache may be associated with focal neurological deficit. [1][2][3] Imaging study may show presence of subarachnoid hemorrhage, intracerebral hematoma, intraventricular hemorrhage, obstructing hydrocephalus, or in late stage may show developing infarct due to vasospasm. However ruptured intracranial aneurysm associated with parasellar mass is uncommon, although cases of incidental aneurysm with intracranial tumor like pituitary adenoma, meningioma, are reported. However, ruptured internal carotid artery aneurysm with metastatic mass lesion is extremely uncommon. Pre-operative knowledge of aneurysms coexisting with sellar mass may help in planning special strategy to deal both lesions simultaneously.

Case Illustration

A-65-year old man was admitted to Department of neurosurgery at neuroscience centre of All India Institute of Medical sciences. He experienced sudden onset very severe headache in left frontal and temporal region associated with vomiting. Next morning, he noticed diminution of vision involving left eye along with drooping of left eyelid. However, he had persistent headache

but intensity has markedly reduced with progressive worsening of drooping of left upper eyelids along with painful progressive ophthalmoplegia. On admission, his supine BP was 130/70. Positive neurological findings included with left eye, he was able to count finger at three feet, while, and on right side was 6/6. Fundi revealed normal retinal arteries and normal appearance of fundi. His extraocular movement of the left eye -ball was completely absent. Routine hemogram and serum biochemistry was normal. Endocrine study revealed serum cortisol (8 am) level $0.8 \mu\text{g} / \text{dl}$ (N 5 -18) TSH- $1.8 \mu\text{IU} / \text{dl}$ (N 0.4- 4.6), T₃ $1.3 \text{pg} / \text{ml}$ (N 1.6-3.4), T₄ – $6.4 \text{ng} / \text{dl}$ (N 0.7- 1.8). As serum cortisol level was low, so corticosteroid replacement was started.

X-ray chest and electrocardiography were normal. An X-ray skull revealed sellar enlargement with erosion of anterior clinoid process. CT scan of cranium revealed sellar, suprasellar lesion with left parasellar extension, showing enhancement with contrast agent. A 4-vessel digital subtraction angiographic study was done for evaluation, which revealed left internal carotid artery aneurysm which was directed superiorly.

Through a left pterional osteoplastic flap craniotomy after eighth day of ictus. There was mass filling up left parasellar and suprasellar region, gross decompression was also done, with intra-operative diagnosis of pituitary adenoma was made. In the postoperative

period, he made good recovery. Just prior to discharge from hospital, a repeat angiography was carried out, revealed well clipped aneurysm to look for completeness of aneurysm clipping. He was discharged from hospital on sixth post-operative day. However, the histopathology turns out to be poorly differentiated metastatic adenocarcinoma.

Discussion

The metastatic mass lesion accounts for only 1% of the tumors located in the sellar-parasellar regions, for cases, who underwent transsphenoidal surgery [1]. Breast and lung cancer are among the most common sites of primary malignant tumors to sellar regions [3] other includes gastrointestinal tract, prostate, kidney, thyroid, and pancreas. The important routes of metastasis to the pituitary gland include hematogenous or direct invasion through the skull base. The hematogenous route is the principal pathway of metastasis and lymphatic system is absent in the brain. The most commonly involved part of the pituitary gland in the order of decreasing frequency is posterior lobe, anterior hypophysis, both the anterior and posterior hypophysis, and the stalk[4]

Intracranial aneurysms commonly occur in the region of Circle of Willis, are often saccular or arteriosclerotic in origin. Although saccular aneurysms can occur in peripheral branches of the circle of Willis, but it is very rare. Aetiology of aneurysms occurring in the peripheral branches includes, in the decreasing order of frequency are mycotic, post-traumatic or secondary to tumour and Moya-Mmoya diseases. Usually the

occurrence of intracranial aneurysm association with intracranial brain tumours or secondary's are extremely uncommon [1][2][3][4][5][6][7][8]

Very rarely incidental aneurysm can be picked up while investigation of sellar mass and few cases of intracranial aneurysm with associated with pituitary adenoma is reported. Such association of intracranial aneurysm with tumor significantly increases the risk of inadvertent rupture during the intraoperative phase; surgery is attempted utilizing either trans-sphenoidal decompression or endoscopic approach.

The commonest presenting symptom in patients with sellar metastases is central diabetes insipidus, followed by anterior hypopituitarism, visual loss, [9][10]

However, diagnose of sellar metastasis is difficult because symptoms are mostly nonspecific and the radiological differences from primary tumors are nonspecific . [12][13][14][15] Imaging study play an important role in the diagnosis of sellar parasellar region, besides clinical examination and hormonal assessment. CT scan head can show presence of subarachnoid hemorrhage, intracerebral hematoma, intraventricular hemorrhage, obstructing hydrocephalus, obstructing hydrocephalus, or in late stage may show developing infarct due to vasospasm. However ruptured intracranial aneurysm associated with parasellar mass further MRI better delineates soft tissues and common lesion in sellar region needs exclusions are meningioma, pituitary adenoma and vascular mass. MRI shows presence of sellar suprasellar mass, showing

contrast enhancement, may be associated with areas of hemorrhage or necrosis representing previous pituitary apoplexy. Meningiomas are solid, with occasional cyst on the edge or associated with peritumoral arachnoid cap, usually show uniform contrast enhancement, In the coronal section images can show a compressed normal pituitary gland can be seen separately from tumour in cases of meningioma at the bottom of the sella turcica and epicenter of mass lies above the sella

A diagnostic MRI sign of flow voids is 100% specific for aneurysms as described by Teng et al. with the sensitivity of 88%, on T1-weighted and T2-weighted imaging sequences[10] however Olsen et al. reported only 80% giant aneurysms shows sign of blood flow in the aneurysm sac. So MRI may fail to detect aneurysm if size of aneurysm being smaller[11]

In , MRI scan, T1W image may show evidence of hypointense signal representing flow void caused by as rapidly flow blood in a carotid aneurysm. Cases of thrombosed aneurysm, DSA study can be very helpful in arriving at diagnosis, delineating shape, size, direction of fundus, vessel originating from aneurysm, relation with adjoining branches of artery, presence of teat.

Regarding management of such co-existing aneurysm with secondary is debated is debated and controversial and depends on size of aneurysm, location on anterior or posterior circulation, proximity to sella or , size of sellar mass adenoma, status of adenoma, volume of adenoma, associated neurological manifestation, visual symptom, extension into multiple cranial fossa or multi-compartmental

and general status of cases. [16][17] [18][19] Although differentiating whether a lesion of interest is primary or secondary is very vital in planning treatment modality for such cases and diagnosis mainly rest on histopathological and immunohistochemistry. If a tumor in the pituitary gland is confirmed to be metastatic, local tumor control is planned to relieve symptoms, and the overall prognosis depends on the site of the primary malignancy[9][10]

Yang et al reported a case of known surgically resected gastric carcinoma in a 57-year-old woman with internal carotid aneurysm developing secondary in the sella. [2] She presented with oculomotor paralysis, postorbital pain, and hypopituitarism as onset symptoms. She had a history of the surgical removal of gastric cancer CT angiography of the intracranial artery revealed an ICA of at the inner edge of the siphon segment. Magnetic resonance imaging and single-photon emission study revealed recurrent sellar mass with intracranial and multiple metastases of bone. He underwent subtotal removal followed by chemotherapy and radiotherapy. [2]

With increasing usage of endovascular approach, aneurysm can be coiled and sellar mass can be resected using microsurgical or endoscopic approach separately, however if aneurysm lying in close proximity to mass can be managed in a single setting with clipping of aneurysm and adenoma resection.[20][21][22] However, approach should be individualized and tailor made after analyzing imaging study. However, high degree of suspicion of associated aneurysm is

must be made, if MRI shows some atypical features and DSA must be carried out.

Conclusion

In cases of suspected subarachnoid hemorrhage following aneurysmal rupture, and imaging study showing sellar mass possibility of primary as well as well as metastatic lesion must be considered. Ruptured aneurysm co-existing with metastatic sellar mass with unknown primary is previously unreported and constitute a rare but difficult to manage association, which has not received proper attention in the literature. As awareness about coexisting aneurysm can aid in proper planning to deal both associated pathology in safest way. if surgery is attempted. However, precise knowledge can avoid injury, if strategy can be planned to deal both lesion in a single operative session.

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