CASE REPORT

A Case of Aberrant Regeneration of Oculomotor Nerve Following Vasculitic Infarct in A Case of Tubercular Meningitis in A Young Female

ABSTRACT

The oculomotor nerve supplies innervation to the medial rectus, inferior rectus, superior rectus, inferior oblique, levator palpebrae superioris, and the parasympathetic supply to the sphincter of the pupil and the ciliary ganglion. Any disruption to the continuity of the nerve anywhere along the course leads to aberrancy or misdirection of the fibers during the periods of regeneration which may be variable leading to misfiring or paradoxical contraction of the muscles supplied by the nerve which is termed as aberrant regeneration or synkinesis of the oculomotor nerve. It's been well documented so far that trauma, tumor, and aneurysms are the most common causes implicated in the process of aberrancy. Synkinesis can affect the lids, pupil, and the palpebral fissure. We report one such case where aberrant regeneration occurred during recovery from a pontine infarct with all classical signs of synkinesis in a young lady with tubercular meningitis. We would like to highlight in the present case report the diverse etiologies, pathogenesis of aberrant regeneration of oculomotor nerve, and likelihood of infarct being one of the causes and various treatment modalities available for the same. All signs of aberrant regeneration due to an uncommon ischemic lesion have been photographically well documented in this case report.

Key words: Neurology, Neuro-ophthalmology, Ophthalmology, Tuberculosis

CASE HISTORY

A 15-year-old girl presented to ophthalmology OPD with a history of the left eye remaining open when looking down for the past 8 months. She also complained of binocular horizontal diplopia in up and down gaze. She was diagnosed with tubercular meningitis (for her symptoms of fever, headache, vomiting, backache, lower limb paresthesia with a history of inability to walk, and seizures) with cerebellar involvement with the left eye pupil involving complete third nerve palsy 8 months ago. She was receiving antitubercular therapy (Category 2) planned for 1 year. Contrast-enhanced CT scan at that time showed few disc and ring T2 hypointense enhancing lesions with surrounding vasculogenic edema in the right parietal post central cortex, bilateral cerebellar hemisphere, and inferior vermis with subtle blooming on GRE seen in inferior vermis lesion possibly calcification suggestive of infective etiology and an acute non-hemorrhagic infarct most likely vasculitis in pontine region, Figure 1.

On examination, her unaided visual acuity recorded 20/20, N6 in both eyes. Ocular motility examination was normal in the right eye whereas evaluation revealed –3 limitation of elevation and –2 limitation of depression in the left eye Figure 2. Color vision testing with Ishihara chart was normal in both eyes. Signs of aberrant regeneration such as pseudo von Graefe's signs (adduction due to contraction of levator palpebrae muscle) and pseudo Duane's sign (adduction of the eye on attempted upward or downward gaze) were present. Pupil in the left eye was mid-dilated sluggish reaction to light. Pupillary constriction was noted and measured with a pupil scale when eye adducted and converged, that is, pseudo Argyll Robertson sign present, Figure 3. Fundus examination was unremarkable in both eyes.

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DISCUSSION

Aberrant regeneration is a very common phenomenon observed with diseases of the third nerve. The exact mechanism is not known but it's been said the regenerating axons from the site of injury also innervate other muscles apart from the ones they are originally supplying leading to cocontraction of multiple muscles. Aberrant regeneration can be primary or secondary. Aberrant regeneration occurs in about 15% of the cases following the third nerve insult.[1] Primary aberrant regeneration is secondary to intracavernous lesions such as meningioma, schwannoma, or aneurysm where the synkinesis occurs without preceding acute nerve paralysis and is due to

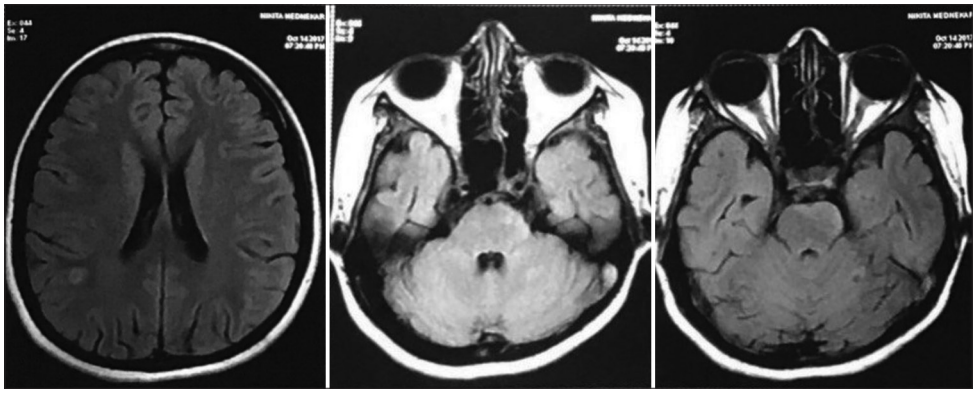

Figure 1: CT brain showing ring-enhancing lesion with pontine infarct

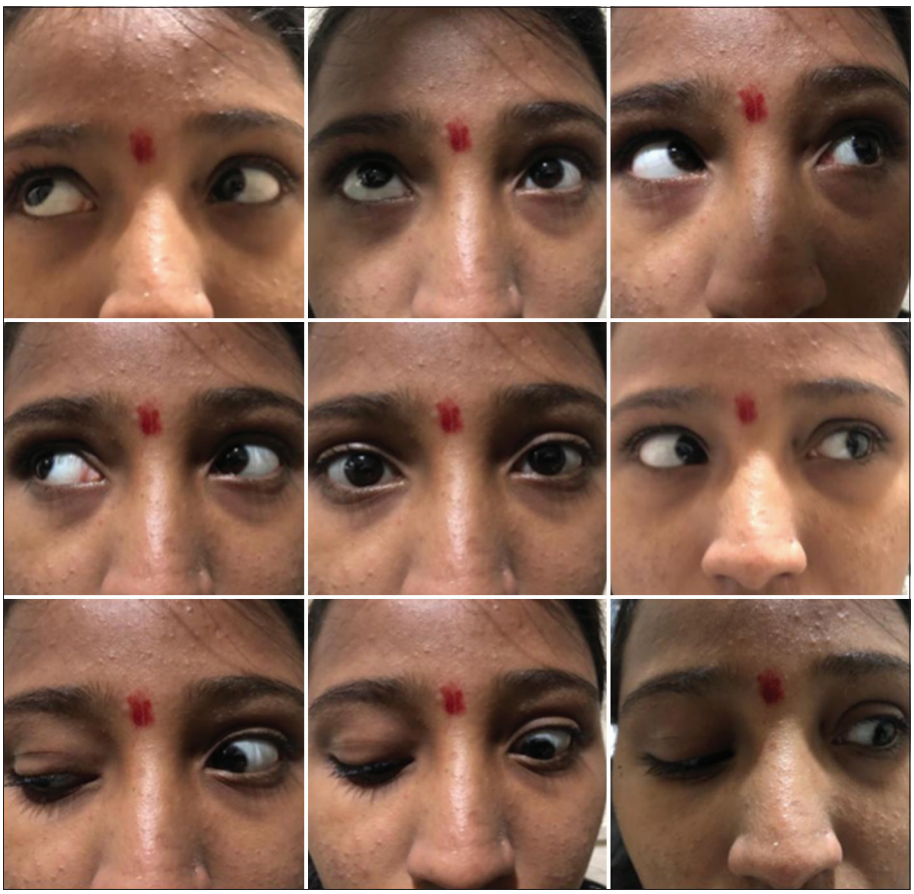

Figure 2: 9 Gaze photo showing –3 limitation of elevation and –2 limitation of depression in left eye with lid retraction on downward gazes

chronic nerve compression.[2] In cases of secondary aberrant regeneration, synkinesis develops after a variable period of several weeks or months when the ptosis and other extraocular movements start recovering.[3] The uncommon causes of synkinesis are ophthalmoplegic migraine,[4,5] Tolosa–Hunt syndrome,[5] and Miller Fisher syndrome.[5]

According to Seddon's classification,[6] aberrancy does not occur in the first- and second-degree nerve injuries. It's more common in the third degree or neurotmesis where the endoneurium is disrupted. Microvascular ischemic damage to the oculomotor nerve due to small vessel disease as in diabetes rarely is responsible for synkinesis, but there are few case reports following the same.[3] Walsh has reported cases of aberrant regeneration following ischemic insult.[3] The most common clinical signs of oculomotor synkinesis consist of elevation of the upper eyelid on attempted downward gaze

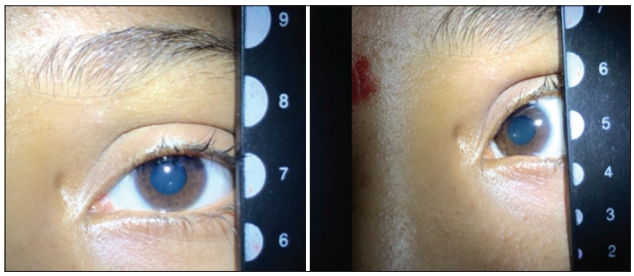

Figure 3: Pseudo Argyll Robertson sign

(pseudo von Graefe's sign) or adduction due to contraction of levator muscle, adduction of the eye on attempted upward or downward gaze (pseudo Duane's sign), and constriction of the pupil on attempted adduction (pseudo Argyll Robertson's sign).

Treatment strategies vary depending on individual clinical manifestations in each patient. Addressal of the cause is most important as is endovascular treatment of her aneurysm using stent-assisted coiling in cases of aneurysms.[7] There is a case report of performing squint surgery in other eye to improve ptosis and signs of aberrant regeneration in the affected eye.[8] Patching, prism therapy, or strabismus surgery may be needed for symptomatic patients but the aberrancy needs to be taken into consideration for surgical planning.

Since our patient was already investigated and treated for the cause, we deferred neuroimaging. Our patient is symptomatic in vertical gaze and more so in down gaze which is mainly essential to carry out daily activities we advised her to have a prism trial to determine if she could fuse in down gaze. The patient was able to achieve single vision, she was given the options of wearing prisms in bifocals or also an alternative surgical procedure of undergoing the right eye inferior rectus faden to overcome the diplopia and achieve a binocular single field of vision depending on the intraoperative forced duction/force generation tests.

CONCLUSION

An acute infarct is one of the several causes of aberrant regeneration of oculomotor nerve which can be easily missed if not considered as one of the differential diagnosis. Neuroimaging is mandatory in all cases of aberrancy to identify the etiology. The treatment in such cases is individualized.

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How to cite this article: Pagad HS, Jain AJ, Shanbhag NU. A Case of Aberrant Regeneration of Oculomotor Nerve Following Vasculitic Infarct in A Case of Tubercular Meningitis in A Young Female. Bombay Hosp J 2021;63(1):31-33.

Source of support: Nil, **Conflicts of interest:** None