Brain Neurorehabil. 2021 Jul;14(2):e17
https://doi.org/10.12786/bn.2021.14.e17
pISSN 1976-8753·eISSN 2383-9910

bnr
Brain & NeuroRehabilitation

Case Report

Bilateral Cerebral Ptosis in a Patient with Subdural Hemorrhage: a Case Report

Ji Hye Kang, Sunha Park, Young-Ah Choi

Revised: Nov 21, 2020
Revised: Mar 19, 2021
Accepted: Mar 28, 2021

Correspondence to
Young-Ah Choi
Department of Rehabilitation Medicine, Incheon St. Mary's Hospital, College of Medicine, The Catholic University of Korea, 56 Dongsu-ro, Bupyeong-gu, Incheon 21431, Korea.
E-mail: simple16@catholic.ac.kr

HIGHLIGHTS

- We describe a patient with bilateral complete ptosis following subdural hemorrhage.
- Interestingly, the patient had no parenchymal lesion.
- The patient made a full recovery from the cerebral ptosis after several weeks.

Brain Neurorehabil. 2021 Jul;14(2):e17
https://doi.org/10.12786/bn.2021.14.e17
pISSN 1976-8753·eISSN 2383-9910

bnr
Brain & NeuroRehabilitation

Case Report

Check for updates

Bilateral Cerebral Ptosis in a Patient with Subdural Hemorrhage: a Case Report

Ji Hye Kang, Sunha Park, Young-Ah Choi

Department of Rehabilitation Medicine, Incheon St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Incheon, Korea

OPEN ACCESS

Revised: Nov 21, 2020
Revised: Mar 19, 2021
Accepted: Mar 28, 2021

Correspondence to
Young-Ah Choi
Department of Rehabilitation Medicine, Incheon St. Mary's Hospital, College of Medicine, The Catholic University of Korea, 56 Dongsu-ro, Bupyeong-gu, Incheon 21431, Korea.
E-mail: simple16@catholic.ac.kr

ORCID iDs
Ji Hye Kang
https://orcid.org/0000-0003-0587-3153
Sunha Park
https://orcid.org/0000-0001-5571-8982
Young-Ah Choi
https://orcid.org/0000-0003-3053-8525

Conflict of Interest
The authors have no potential conflicts of interest to disclose.

ABSTRACT

Although cerebral ptosis is rare, it is commonly associated with unilateral right cerebral hemisphere lesions. We report a case of a 79-year-old woman who presented with bilateral complete ptosis after a traumatic right fronto-temporo-parietal subdural hemorrhage (SDH). Bilateral ptosis was the primary manifestation of the acute right SDH, and the patient had no parenchymal lesion. Her prognosis was good, and she made a complete recovery. Right hemispheric hypoperfusion, as demonstrated on brain perfusion single-photon emission computed tomography, implied that the lateralization of eyelid control was in the right hemisphere, in line with previous reports.

Keywords: Cerebral Ptosis; Single-photon Emission Computerized Tomography; Subdural Hemorrhage

INTRODUCTION

Ptosis is a condition where the upper eyelid falls down, making it difficult to open the eye and narrowing the margin of the upper eyelid [1]. The differential diagnosis of acquired ptosis can be challenging due to multiple possible etiologies, including supranuclear lesions, oculomotor complex lesions, oculosympathetic lesions, neuromuscular junction dysfunction, neuromuscular disease, and mechanical lid abnormalities [2]. Cerebral ptosis (CP) refers to weakness of the levator palpebrae superioris muscle attributable to a cerebral hemispheric lesion and not the brainstem, oculomotor nerve, or oculosympathetic fibers [1]. CP can occur unilaterally or bilaterally. A contralateral cerebral hemispheric lesion, such as a middle cerebral artery infarction, tumor, or arteriovenous malformation, can cause a unilateral supranuclear ptosis. On the contrary, bilateral CP may occur with unilateral or bilateral hemispheric lesions [3].

Excluding emergency situations is crucial while diagnosing CP, since ptosis can be a warning sign of severe medical problems; for example, bilateral complete ptosis can be confused with a state of decreased consciousness. In the rare case of massive infarction of the right hemisphere, bilateral CP is also a sign of impending herniation [4].

Although CP is considered rare, bilateral CP caused by traumatic brain injury without a cerebral parenchymal lesion has never been reported. Herein, we report a case of bilateral CP developed after acute right subdural hemorrhage (SDH). The patient has provided informed consent for the publication of this case.

CASE REPORT

A 79-year-old woman with hypertension and diabetes mellitus was hospitalized for mild left hemiparesis and bilateral CP that developed after traumatic brain injury (TBI) due to falling down in her room, accompanied with loss of consciousness. Initially, her glasgow coma scale was 15 with no evidence of post-traumatic amnesia. Computed tomography (CT) of the brain revealed a traumatic right fronto-temporo-parietal SDH without any skull fracture or other structural abnormalities that could damage the pathway of the oculomotor nerve. No evidence of any other parenchymal or midbrain lesions was observed by magnetic resonance imaging (MRI; Fig. 1). Mild midline herniation was observed on performing brain imaging studies. She underwent an urgent craniotomy for the removal of the SDH and received post-operative care in the intensive care unit for seven days. For additional neuroimaging information, brain perfusion Tc-99m EDC single-photon emission CT (SPECT) was performed to evaluate brain metabolic activity. The brain perfusion SPECT revealed hypoperfusion in the right temporal region and right basal ganglia (Fig. 2).

Neurologic examination performed after surgery revealed that the patient had an alert mental status and was obeying complex commands. Initially, the manual muscle test (MMT) [5] grade of the right side was 3 and that of the left side was 2, and she was completely dependent for daily life activities. The cranial nerve examination revealed no abnormalities; gaze deviation or visual field loss were not found, and the pupils were symmetric, reactive, and normal-sized. Additionally, no extra-ocular movement (EOM) limitation or decrease in visual acuity was observed. However, she had complete bilateral ptosis even when attempting to open her eyes. Contraction of the levator palpebrae superioris muscle was not visible, and instead, compensatory contraction of the frontalis muscle was observed. Furthermore, involuntary eye opening was also impaired. The results of facial nerve conduction studies, electromyography, and blink reflex studies to evaluate the temporal and zygomatic branches of the facial nerve were normal.

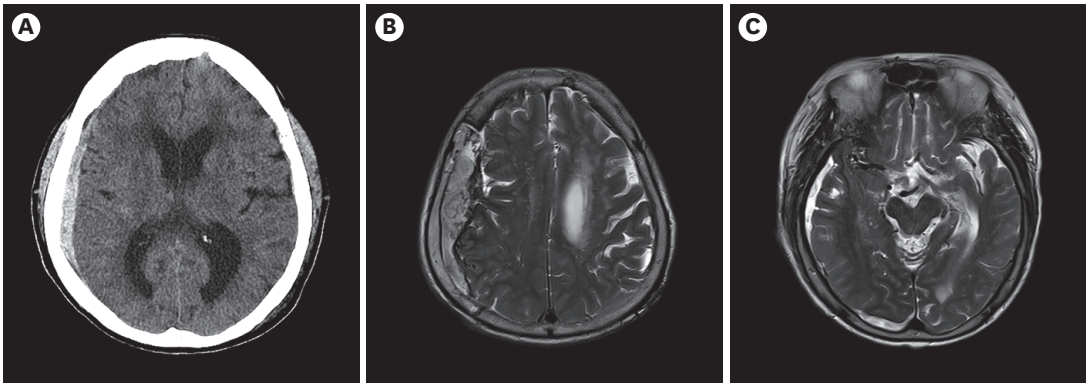

Fig. 1. Brain CT and MRI demonstrate a right fronto-temporo-parietal subdural hematoma. (A) Initial brain CT (axial). (B) Brain T2-weighted MRI (axial) without parenchymal lesion. (C) Brain T2-weighted MRI (axial) without midbrain lesion.
CT, computed tomography; MRI, magnetic resonance imaging.

https://doi.org/10.12786/bn.2021.14.e17

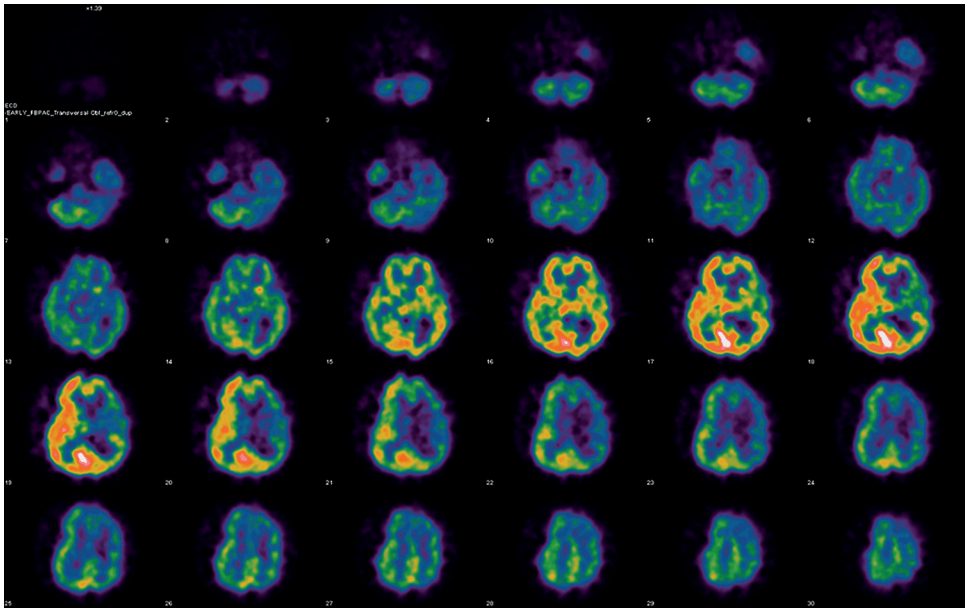

Fig. 2. Brain perfusion Tc-99m EDC single-photon emission computed tomography demonstrates hypoperfusion in the left frontal and right temporal regions, and the right basal ganglia.

Although she had difficulty in participating in the rehabilitation programs due to complete bilateral ptosis, she received rehabilitation training via activities on a mat with the assistance of physical therapists and an occupational therapist. To improve the effectiveness of the rehabilitation, double eyelid tapes for upward fixation of the upper eyelid and eyelid glue, which is an adhesive that fixes the eyelids to the supraorbital structures, were temporarily applied during rehabilitation treatment.

Bilateral ptosis tends to improve after three weeks following the onset of TBI. Three weeks after the onset of TBI, the vertical length of the palpebral fissure was measured as 4 mm bilaterally, and the bilateral upper lid margin-corneal light reflex distance was 0 mm. The bilateral levator excursion amplitudes from downgaze to upgaze were measured as 6 mm. The bilateral ptosis gradually improved with intensive rehabilitation. Six weeks after onset, the palpebral aperture measured 7 mm bilaterally, the superior margin-reflex distance was 3 mm bilaterally, and the levator excursion amplitudes from downgaze to upgaze measured 11 mm bilaterally.

At the time of discharge, the patient's bilateral ptosis was completely resolved. Her muscle strength also improved to MMT grade 5 on both the sides, and she could walk and perform daily life activities independently.

DISCUSSION

Bilateral CP is a rare and challenging condition to diagnose. A majority of the literature has described it in association with right hemisphere vascular damage, such as middle cerebral artery stroke [6,7]. SDH is a collection of blood between the dura and the arachnoid membranes, usually induced by head trauma caused due to motor vehicle accidents, falls, and assaults [8]. To our knowledge, this is the first case report to state that acute traumatic SDH on the right side could result in bilateral CP.

https://doi.org/10.12786/bn.2021.14.e17

For the diagnosis of ptosis, history taking and physical examination is important [9,10]. In this case, we could exclude ptosis caused by orbital lesions initially by history taking. Additionally, myogenic ptosis could be ruled out from normal findings of electromyography and acetylcholine receptor antibody test. Thereby, neurogenic ptosis is the most likely cause. Upon physical examination, the pupil, EOM, and cranial nerve were found to be normal. The upper eyelid muscles and their innervations did not correspond to the general differential diagnosis of ptosis caused by the oculomotor nerve, oculosympathetic neurons, and the facial nerve. Excluding oculomotor nerve palsy is important when the levator palpebrae superioris muscle exhibits abnormal findings. Oculomotor nerve palsy can be caused by problems in the pathway from the oculomotor nucleus in the midbrain to the extraocular muscles, eyelids, and pupils. It passes through the midbrain, the subarachnoid space, the lateral wall of the cavernous sinus, and is divided into superior/inferior branches to the ocular area of extraocular muscles, eyelids, and pupils [11]. Since no structural abnormality invading the pathway was observed in brain imaging studies, the probability of oculomotor nerve palsy is unlikely. Based on this, the ptosis was presumed to be CP, caused by right hemisphere brain activity reduction. CP is very rare, and its mechanism is not clearly identified; hence, it is important to perform relevant differential diagnosis for it.

The criteria for the diagnosis of CP have not yet been established. Manconi et al. [12] proposed inclusion and exclusion criteria. The reported inclusion criteria were as follows: 1) sudden bilateral lid drop within 48 hours of stroke, 2) voluntary, spontaneous, and automatic impairment in eyelid opening, 3) preserved voluntary frontalis muscle contraction, and 4) neuroradiological evidence of supratentorial ischemic or hemorrhagic damage. The exclusion criteria were presented together to distinguish the following causes of ptosis: apraxia of lid opening (ALO), impaired consciousness, blepharospasm, intrinsic oculomotor dysfunction, neuromuscular disease, brain stem dysfunction, and subtentorial lesions. In this case, ALO and blepharospasm should be differentially diagnosed. ALO is a condition of abnormal motor control of the eyelid causing difficulty in initiating lid elevation. It is related to lesions of the right cerebral hemisphere or bilateral hemispheric lesions, and also occurs with extrapyramidal disorders like progressive supranuclear palsy or Parkinson's disease [13,14]. Both voluntary and involuntary eye opening were impaired in this case, and manually lifting the eyelid was also ineffective. Furthermore, blepharospasm was also excluded, as there were no orbicularis oculi muscle contractions to prevent the eyelids from opening. Since the location of the SDH in this case was supratentorial, it partially satisfies the fourth inclusion criteria, although the notable difference in this case was that an extra-axial lesion rather than an ischemic or hemorrhagic lesion, caused the CP.

Although the mechanisms and clinical significance of CP remain uncertain, experiments in both, animals and humans have shown that opening of both eyelids occurs upon stimulation of the frontal and occipital lobes [15,16]. Clinically, it has been hypothesized that eyelid motor control might be lateralized to the right hemisphere in complete CP [6,7,17]. However, the specific mechanism is not yet fully elucidated, and the above hypothesis is still controversial [4]. SPECT was performed to confirm brain activity because the patient's CP persisted. SDH can reduce brain activity by increasing the intracranial pressure and reducing cerebral perfusion pressure and cerebral blood flow. Subdural blood components can also reduce the metabolism of the brain. Baechli et al. [18] showed that in rats with acute subdural hematoma, pathomechanism by blood constituents aggravated brain edema and reduction of glucose metabolism. Schröder et al. [19] introduced two cases and showed that cerebral blood flow, which had decreased due to subdural hematoma, increased again after hematoma

https://doi.org/10.12786/bn.2021.14.e17

removal. Although there was no parenchymal lesion found on imaging, the clinical manifestations combined with the brain SPECT results indicated temporary dysfunction or suppression of the right cerebral hemisphere. This finding is in line with previous reports of dominance of the right hemisphere in lid control [6,20]. The hypoperfusion in the right basal ganglia on the brain SPECT in this case may be a clue to the mechanism of bilateral CP. The basal ganglia are hypothesized to be involved in the regulation of eye opening in the supplementary motor area via the thalamus [21]. Based on this, it can be speculated that the decreased brain activity in the right hemisphere caused bilateral CP by weakening the levator palpebrae superioris muscle in this patient.

The prognosis of CP is variable and depends on the lesion and etiology [14]. Manconi et al. [12] reviewed literature on 75 CP patients, reporting an improvement in ptosis in 70.9%, with an average recovery time of 7.5 days. In the case of our patient, not even slight improvement was observed until three weeks after the onset, and the patient recovered completely only six weeks after the onset. Although the recovery time for the patient in the present case was much longer than average, the prognosis of CP due to SDH in the right hemisphere can be considered to be good.

In conclusion, bilateral CP is usually associated with right hemisphere infarction, and early diagnosis is urgent since acute bilateral CP is usually associated with impending cerebral herniation. However, it can also be the primary manifestation of acute right SDH without a parenchymal lesion, where the prognosis of CP is good. In this case, the patient without abnormal findings in brain MRI was diagnosed with CP using SPECT, to confirm decreased brain activity, and by excluding other causes through several tests.

REFERENCES

1. Caplan LR. Ptosis. J Neurol Neurosurg Psychiatry 1974;37:1-7.
PUBMED | CROSSREF
2. Lee AG, Brazis PW. Clinical pathways in neuro-ophthalmology: an evidence-based approach. 2nd ed. New York, NY: Thieme Medical Publishing; 2003.
3. Nutt JG. Lid abnormalities secondary to cerebral hemisphere lesions. Ann Neurol 1977;1:149-151.
PUBMED | CROSSREF
4. Blacker DJ, Wijdicks EF. Delayed complete bilateral ptosis associated with massive infarction of the right hemisphere. Mayo Clin Proc 2003;78:836-839.
PUBMED | CROSSREF
5. Cuthbert SC, Goodheart GJ Jr. On the reliability and validity of manual muscle testing: a literature review. Chiropr Osteopat 2007;15:4.
PUBMED | CROSSREF
6. Averbuch-Heller L, Leigh RJ, Mermelstein V, Zagalsky L, Streifler JY. Ptosis in patients with hemispheric strokes. Neurology 2002;58:620-624.
PUBMED | CROSSREF
7. Averbuch-Heller L, Stahl JS, Remler BF, Leigh RJ. Bilateral ptosis and upgaze palsy with right hemispheric lesions. Ann Neurol 1996;40:465-468.
PUBMED | CROSSREF
8. Bullock MR, Chesnut R, Ghajar J, Gordon D, Hartl R, Newell DW, Servadei F, Walters BC, Wilberger JE; Surgical Management of Traumatic Brain Injury Author Group. Surgical management of acute subdural hematomas. Neurosurgery 2006;58:S16-24.
PUBMED
9. Finsterer J. Ptosis: causes, presentation, and management. Aesthetic Plast Surg 2003;27:193-204.
PUBMED | CROSSREF

10. Díaz-Manera J, Luna S, Roig C. Ocular ptosis: differential diagnosis and treatment. Curr Opin Neurol 2018;31:618-627.
PUBMED | CROSSREF
11. Brazis PW. Localization of lesions of the oculomotor nerve: recent concepts. Mayo Clin Proc 1991;66:1029-1035.
PUBMED | CROSSREF
12. Manconi M, Cesnik E, Casetta I, Govoni V, Ferini-Strambi L, Granieri E. Cerebral blepharoptosis: two new cases, literature review and proposal for diagnostic criteria. Neurol Sci 2006;27:161-165.
PUBMED | CROSSREF
13. Hamedani AG, Gold DR. Eyelid dysfunction in neurodegenerative, neurogenetic, and neurometabolic disease. Front Neurol 2017;8:329.
PUBMED | CROSSREF
14. Dimitriou J, Montoute T, Levivier M, Borruat FX, Diserens K. Bilateral ptosis: lesion in the oculomotor nuclei or supranuclear lesion? NeuroRehabilitation 2015;36:323-327.
PUBMED | CROSSREF
15. Kennard MA, Ectors L. Forced circling in monkeys following lesions of the frontal lobes. J Neurophysiol 1938;1:45-54.
CROSSREF
16. Bender MB, Fulton JF. Functional recovery in ocular muscles of a chimpanzee after section of oculomotor nerve. J Neurophysiol 1938;1:144-151.
CROSSREF
17. Lepore FE. Bilateral cerebral ptosis. Neurology 1987;37:1043-1046.
PUBMED | CROSSREF
18. Baechli H, Behzad M, Schreckenberger M, Buchholz HG, Heimann A, Kempski O, Alessandri B. Blood constituents trigger brain swelling, tissue death, and reduction of glucose metabolism early after acute subdural hematoma in rats. J Cereb Blood Flow Metab 2010;30:576-585.
PUBMED | CROSSREF
19. Schröder ML, Muizelaar JP, Kuta AJ. Documented reversal of global ischemia immediately after removal of an acute subdural hematoma. Report of two cases. J Neurosurg 1994;80:324-327.
PUBMED | CROSSREF
20. Schmidtke K, Büttner-Ennever JA. Nervous control of eyelid function. A review of clinical, experimental and pathological data. Brain 1992;115:227-247.
PUBMED | CROSSREF
21. Costa J, Valls-Solé J, Valldeoriola F, Rumià J, Tolosa E. Motor responses of muscles supplied by cranial nerves to subthalamic nucleus deep brain stimuli. Brain 2007;130:245-255.
PUBMED | CROSSREF