

## Audible blink in carotid-cavernous fistula

A carotid-cavernous fistula (CCF) is an abnormal arteriovenous communication between the carotid artery system and the cavernous sinus. Common ocular manifestations of CCFs include pulsating exophthalmos, conjunctival chemosis, periorbital bruit, diplopia, and ophthalmoplegia.<sup>1</sup> We report a patient with atypical CCF who presented with audible blinks and describe the possible pathogenesis of this phenomenon.

A 54-year-old white female presented with intermittent double vision, eye redness, facial pain, and lid swelling OS with an unusual complaint of audible blinks OS. Her medical history was significant for pituitary hyperplasia status post transsphenoidal resection, stage 0 breast cancer, and a remote history of *Bartonella henselae* neuroretinitis. Medication, family, and social histories were noncontributory.

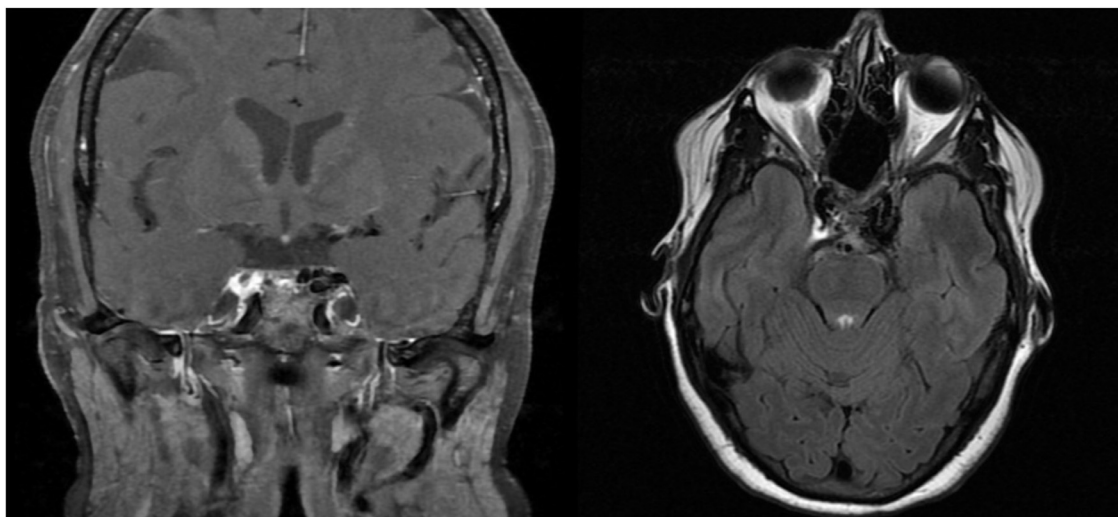
The patient presented initially with headache and progressive bitemporal visual field loss. Cranial magnetic resonance imaging confirmed an enlarged pituitary lesion compressing the optic chiasm. Transsphenoidal resection and biopsy showed focal fibrosis of the adenohypophysis. Postoperative imaging was negative for recurrence, and her symptoms and signs resolved.

Four months later, the patient presented with an unusual audible clicking sound with blinking, intermittent diplopia, dry eye, and lid swelling OS that would decrease during the day and after sleeping upright. Visual acuity was 20/25 OD and 20/40 OS. Motility was full OU without nystagmus. Her monocular horizontal diplopia OS worsened with extreme left gaze only with difficulty realigning eyes returning from the gaze consistent with a preexisting left cranial nerve VI palsy. External examination showed moderate lid

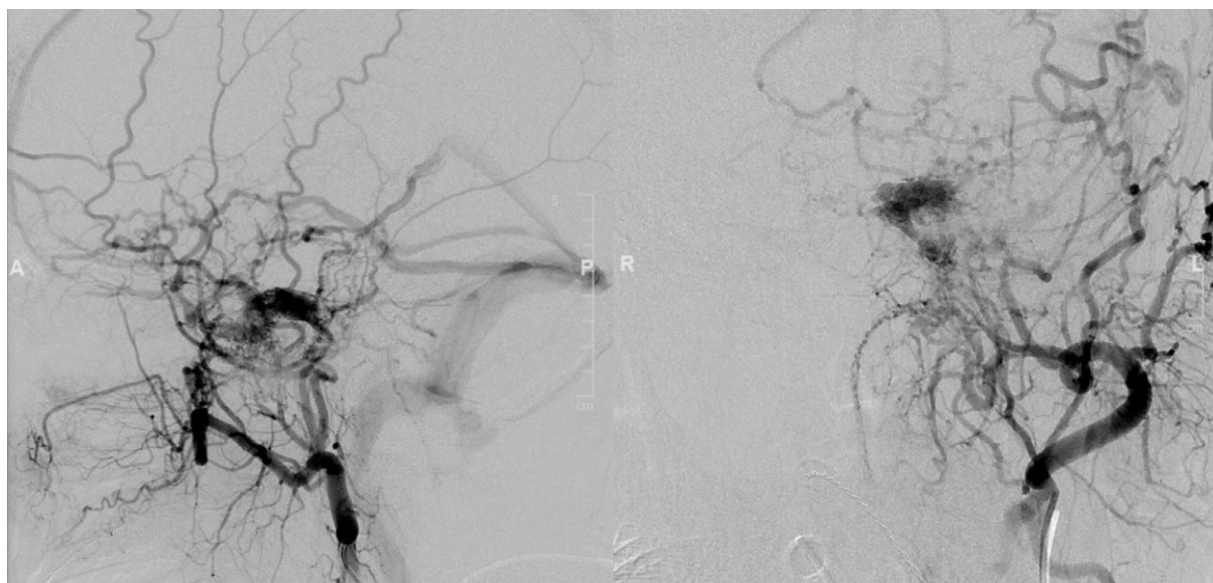
edema, mild ptosis, mild lacrimal gland enlargement, and conjunctival chemosis OS. Slit-lamp biomicroscopy showed a mild nuclear sclerotic cataract consistent with 20/40 vision OS. Because the clicking sounds resolved by the appointment, the patient showed a video recording of her blinking that confirmed the symptom of audible blinks that presented on upward and downward excursions of the eyelid OS without a visible air bubble (Video 1, available online).

Laboratory studies including tuberculosis, syphilis, IgG subclass levels, pituitary hormones, vitamin B<sub>12</sub>, and thyroid function studies were unremarkable. Magnetic resonance imaging of the brain and orbits revealed asymmetric prominence of the left cavernous sinus (Fig. 1). Cerebral angiogram revealed a left-sided Cognard type IV dural CCF and right-sided Cognard type I dural CCF (Fig. 2). The patient underwent coil embolization and complete obliteration of the left CCF, which resolved the lid edema and audible blinks OS. At the follow-up visit, her vision remained stable, but she had a postoperative left abduction deficit with worsened diplopia consistent with left cranial nerve VI palsy.

Direct CCFs have a direct connection between the internal carotid artery and the cavernous sinus, whereas dural CCFs have an indirect connection involving the cavernous sinus and cavernous arterial branches.<sup>2</sup> Cognard classification distinguishes between benign and aggressive types of dural CCFs by cortical venous drainage (CVD), dural sinus drainage, and venous outflow architecture.<sup>3</sup> Ocular manifestations of CCFs include pulsatile exophthalmos, chemosis, periorbital bruit, ophthalmoplegia, elevated intraocular pressure, and exposure keratopathy.<sup>1</sup> If there is a suspicion for CCF, angiography is the accurate diagnostic method to identify the location of a fistula, draining



**Fig. 1**—Coronal fat-suppressed T<sub>1</sub>-weighted postcontrast (A) and axial T<sub>2</sub>-weighted fluid-attenuated inversion recovery (B) magnetic resonance imaging demonstrating a nonspecific asymmetric prominence in the left cavernous sinus.



**Fig. 2—Lateral left (A) and frontal (B) cerebral angiograms reveal a left-sided Cognard type IV dural carotid-cavernous fistula.**

pattern, flow rate, and reflux.<sup>1,3</sup> Endovascular intervention is recommended for CCFs with high-flow fistulas or CVD because of possible vascular complications.<sup>3</sup> Low-risk cases are managed conservatively because some resolve spontaneously.<sup>1,3</sup> However, secondary glaucoma, diplopia, intolerable bruit or headache, exposure keratopathy, or debilitating visual symptoms could suggest an indication for intervention.<sup>1,2</sup>

Our patient had a left-sided Cognard type IV dural CCF, which consisted of CVD and venous ectasia; she also had a right-sided Cognard type I dural CCF, which was benign and showed no signs of CVD.<sup>3</sup> We hypothesize that the CCF produced intermittent lid swelling OS. The audible blinks that improved with her upright posture suggest a gravity-dependent phenomenon. Her ocular symptoms and audible blinks could be the result of increased resistance from the retrograde venous drainage into the ophthalmic and facial veins through retrograde flow from the type IV dural CCF, leading to the intraorbital fluid accumulation.<sup>1</sup> Fluid collection in the dependent supine position and nocturnal lagophthalmos also might be factors in the morning that led to audible blinks and resolved as vascular congestion improved throughout the day. Postoperatively, our patient's audible blinks and lid edema resolved along with resolution of the left CCF. It is not clear why audible blinks are not heard more often, however, in patients with thyroid eye disease or CCF.

Interestingly, the only previously reported cases of audible blinks in the literature were described as a side effect of prostaglandin analogue use and the silent sinus syndrome.<sup>4,5</sup> We speculate that enophthalmos could lead to a negative pressure and air entrapment after eyelid closure, producing a clicking sound on eyelid opening and air bubble "bursting."<sup>4,5</sup> Alternatively, intraorbital fluid rather than air accumulation could have produced the audible blink. To

the best of our knowledge, this is the first case of CCF presenting with audible blinks to be described in the English-language literature.

**Online-only material:** This article includes online-only material. Video 1 can be found on the CJO web site at <http://pubs.nrc-cnrc.gc.ca/cjo/cjo.html>. It is linked to this article in the online contents of the xxx 2022 issue.

## Supplementary materials

Supplementary material associated with this article can be found in the online version at doi:10.1016/j.jcjo.2022.06.003.

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## References

1. Chaudhry IA, Elkhamry SM, Al-Rashed W, Bosley TM. Carotid cavernous fistula: ophthalmological implications. *Middle East Afr J Ophthalmol* 2009;16:57–63.

- 221 2. Barrow DL, Spector RH, Braun IF, Landman JA, Tindall SC,  
222 Tindall GT. Classification and treatment of spontaneous  
223 carotid-cavernous sinus fistulas. *J Neurosurg* 1985;62:248–56.  
224 3. Gandhi D, Chen J, Pearl M, Huang J, Gemmete JJ, Kathuria  
225 S. Intracranial dural arteriovenous fistulas: classification,  
226 imaging findings, and treatment. *AJNR Am J Neuroradiol*  
227 2012;33:1007–13.  
228 4. Abedi F, Chappell A, Craig JE. Audible clicking on blinking:  
229 an adverse effect of topical prostaglandin analogue medica-  
230 tion. *Clin Exp Ophthalmol* 2017;45:304–6.

- 277 5. Dailey RA, Cohen JI. Surgical repair of the silent sinus syn-  
278 drome. *Ophthalmic Plast Reconstr Surg* 1995;11:261–8.  
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### Footnotes and Disclosure

The authors have no proprietary or commercial interest in  
any materials discussed in this correspondence.

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