DOI: 10.5603/oj.100204

Bilateral 6th nerve palsy due to intracranial hypertension after methyl methacrylate cranioplasty: a case report

Taoufik Abdellaoui[1], Sebbata Soundouss[2], Mouzari Yassine[2], Oubaaz Abdelbarre[2]

[1]Department of Ophthalmology, Sidi Mohamed ben Abdellah University, Fes, Morocco
[2]Mohamed-V Military Teaching Hospital, Rabat, Morocco

ABSTRACT

BACKGROUND: Intracranial hypertension after cranioplasty is unusual. We report a case of bilateral 6th nerve palsy secondary to intracranial hypertension that occurred in the immediate aftermath of methyl methacrylate cranioplasty following surgical resection of Ewing's sarcoma of the occipital bone.

CASE PRESENTATION: A 21-year-old woman presented with a gradually enlarging occipital mass, non-painful but cosmetically concerning. Angio scanning revealed a large, vascularized, extracranial mass with malignant features and occipital bone erosion. Surgical excision, including bone resection, identified Ewing's sarcoma. Postoperatively, she developed headaches, blurred vision, and diplopia. Ophthalmological examination revealed swollen optic discs and restricted ocular motility. Imaging ruled out intracranial involvement but showed inflammatory changes at the cranioplasty site. Lumbar puncture confirmed intracranial hypertension secondary to cranioplasty. Treatment with acetazolamide and potassium led to a positive functional outcome.

CONCLUSIONS: This case underscores the need for heightened awareness regarding potential complications associated with cranioplasty, particularly when utilizing materials like methyl methacrylate.

KEYWORDS: intracranial hypertension; cranioplasty; methyl methacrylate; 6th nerve palsy; Ewing's sarcoma

Ophthalmol J 2024; Vol. 9, 149–154

INTRODUCTION

In the intricate realm of neurosurgery, cranioplasty stands as a crucial procedure addressing both structural and aesthetic aspects, but it can introduce a spectrum of potential postoperative complications. This article presents an unusual case of bilateral 6th nerve palsy resulting from intracranial hypertension immediately following methyl methacrylate (MMA) cranioplasty performed after the surgical resection of Ewing's sarcoma in the occipital bone of a 21-year-old woman.

CASE REPORT

A 21-year-old woman presented with a swelling in the occipital region that had been gradually increasing in size for the past year. There was no pain or associated general symptoms; rather, the patient sought medical attention due to cosmetic discomfort. Angio scanning revealed a large extracranial, isodense mass with malignant features measuring 12.5 × 7.5 × 9.5 cm. The mass was vascularized, associated with erosion of the occipital bone, and showed no intracranial extension (Fig. 1).

CORRESPONDING AUTHOR:
Taoufik Abdellaoui, Department of Ophthalmology, Sidi Mohamed ben Abdellah University, Fes, Morocco, Agdal, Rabat, Morocco, 12020;
e-mail: taoufik.abdellaoui@hotmail.com

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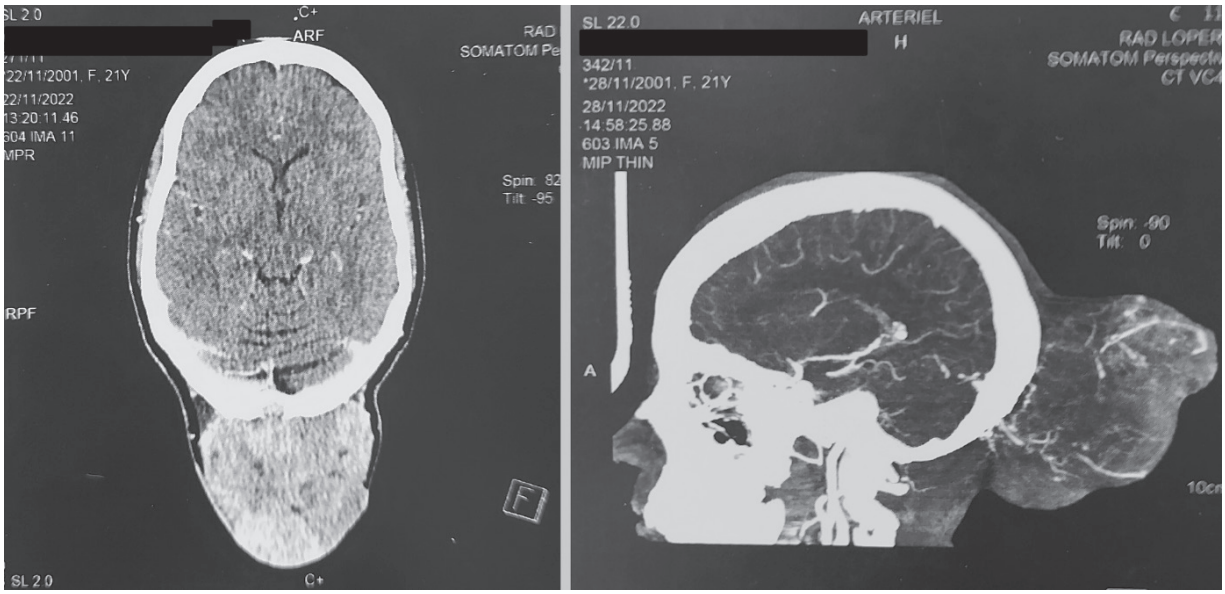

FIGURE 1. Cerebral angioscan: large extracranial mass, measuring 12.5 × 7.5 × 9.5 cm, vascularized, with erosion of the occipital bone, and no intracranial extension

The patient underwent surgical excision of the tumor, with resection of the eroded occipital bone. There was no involvement of the internal aspect of the occipital bone or invasion of the underlying dura mater. Subsequently, a cranioplasty with polymethyl methacrylate was performed. Pathological examination of the operative specimen identified Ewing's sarcoma. Additional radio-chemotherapy was decided upon.

The patient experienced headaches, visual blurring, and diplopia in the immediate postoperative period. Ophthalmological examination revealed, in both eyes, a visual acuity of 9/10, normal anterior segments, and an intraocular pressure of 14 mm Hg. Fundoscopic examination demonstrated bilaterally symmetrical swollen optic discs with blurred margins, without haemorrhages or exudates suggestive of papilledema (Fig. 2). The ocular mo-

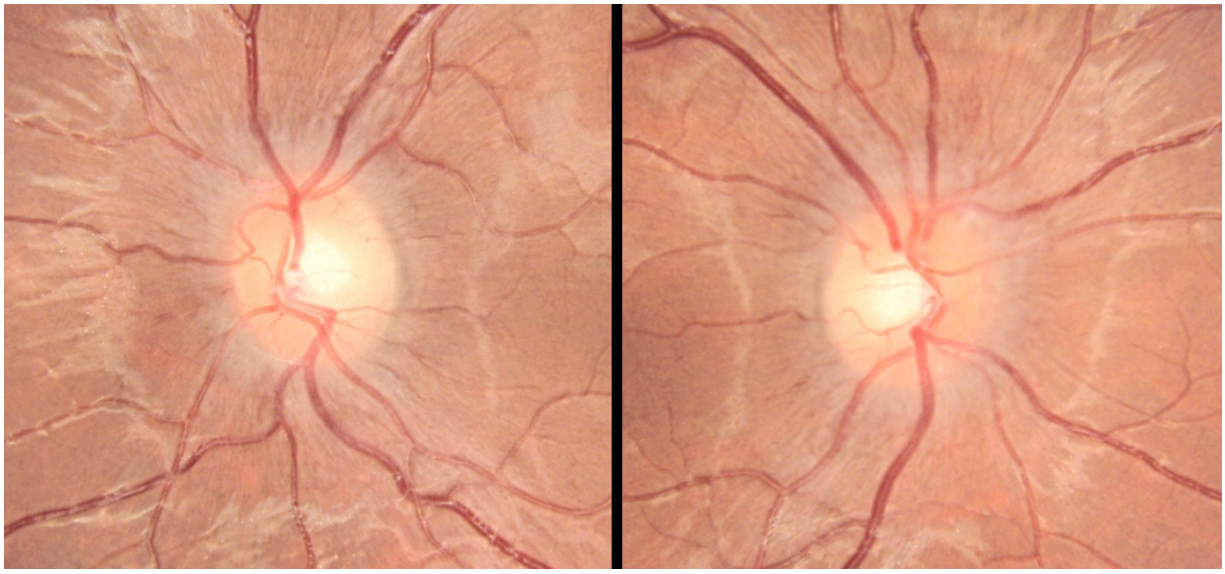

FIGURE 2. Photographs of optic discs: protruding optic discs with blurred edges without haemorrhages or exudates suggestive of stasis papilledema

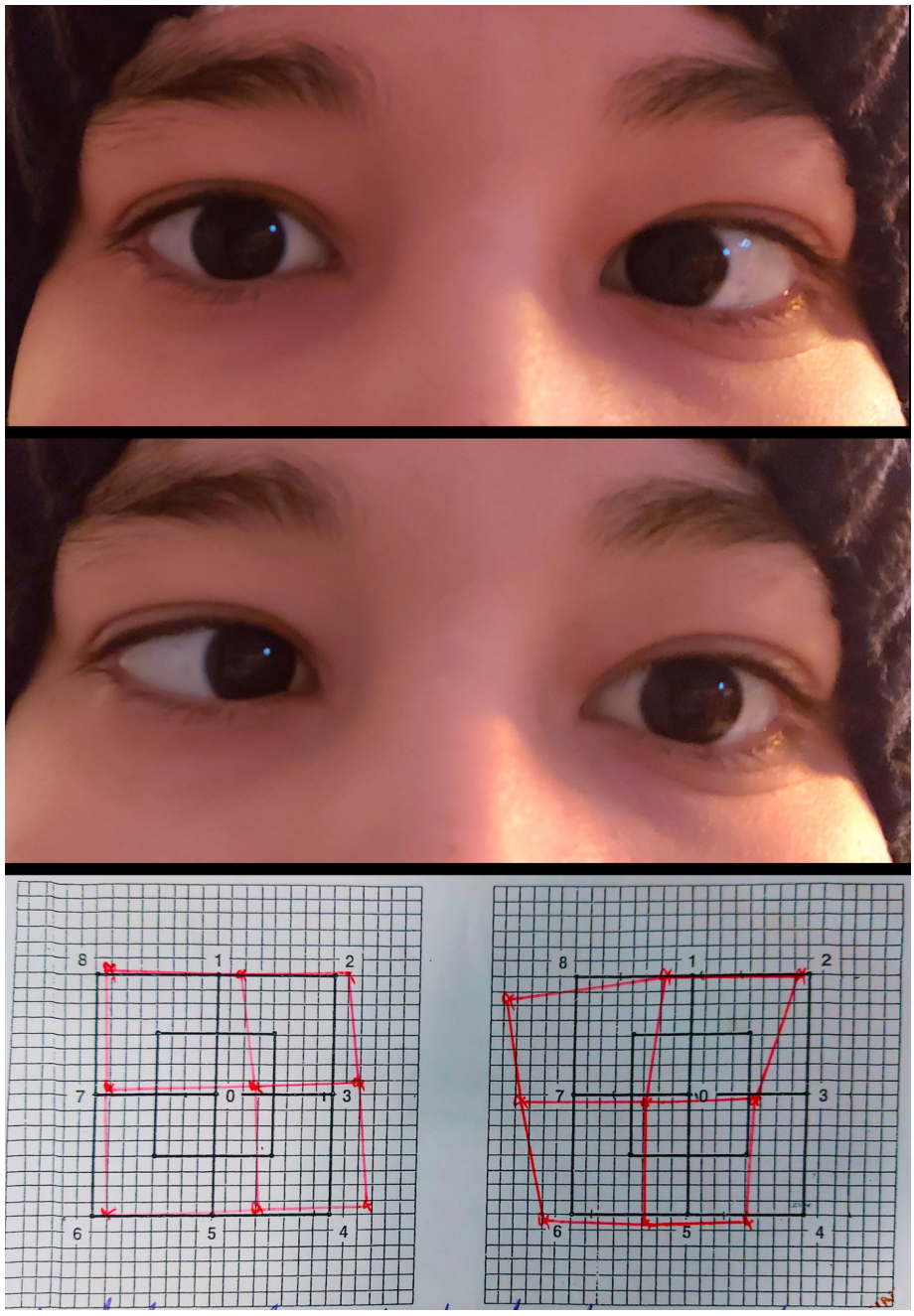

FIGURE 3. Clinical examination and Lancaster test: limitation of external rectus muscles and hyperaction of internal rectus muscles

tility was restricted in the external gaze in both eyes. The Lancaster test revealed a limitation of the external rectus muscles and hyperaction of the internal rectus muscles (Fig. 3).

Computed tomography and magnetic resonance imaging showed inflammatory changes at the cranioplasty site, ruling out intracranial involvement or cerebral thrombophlebitis (Fig. 4). Lumbar puncture indicated an opening pressure of 35 cm H_2O with normal cerebrospinal fluid composition. The diagnosis of intracranial hypertension secondary to cranioplasty was established. The patient was treated with acetazolamide 3 g/day and potassium supplementation, resulting in a good functional recovery (Fig. 5).

DISCUSSION

Ewing sarcoma, with an incidence of 2%, stands as one of the most dreadful cancers. While it could concern people of all ages, it mainly affects children. It is a malignant tumor that primarily

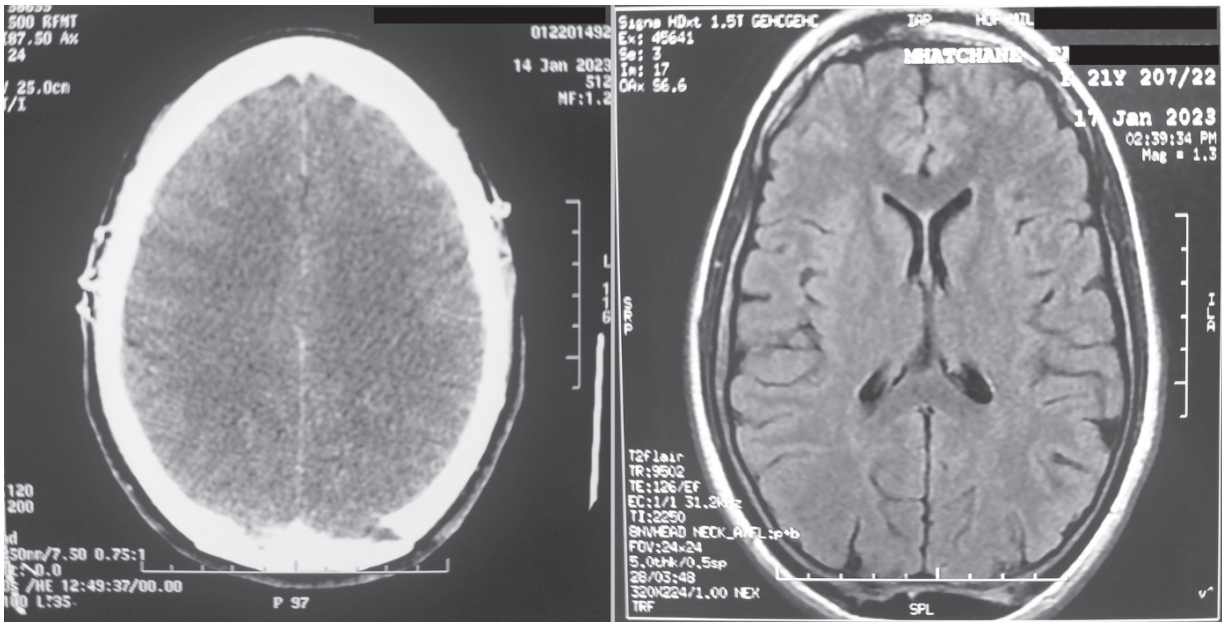

FIGURE 4. Computed tomography and magnetic resonance imaging: inflammatory changes at the cranioplasty site, no intracranial lesions or cerebral thrombophlebitis

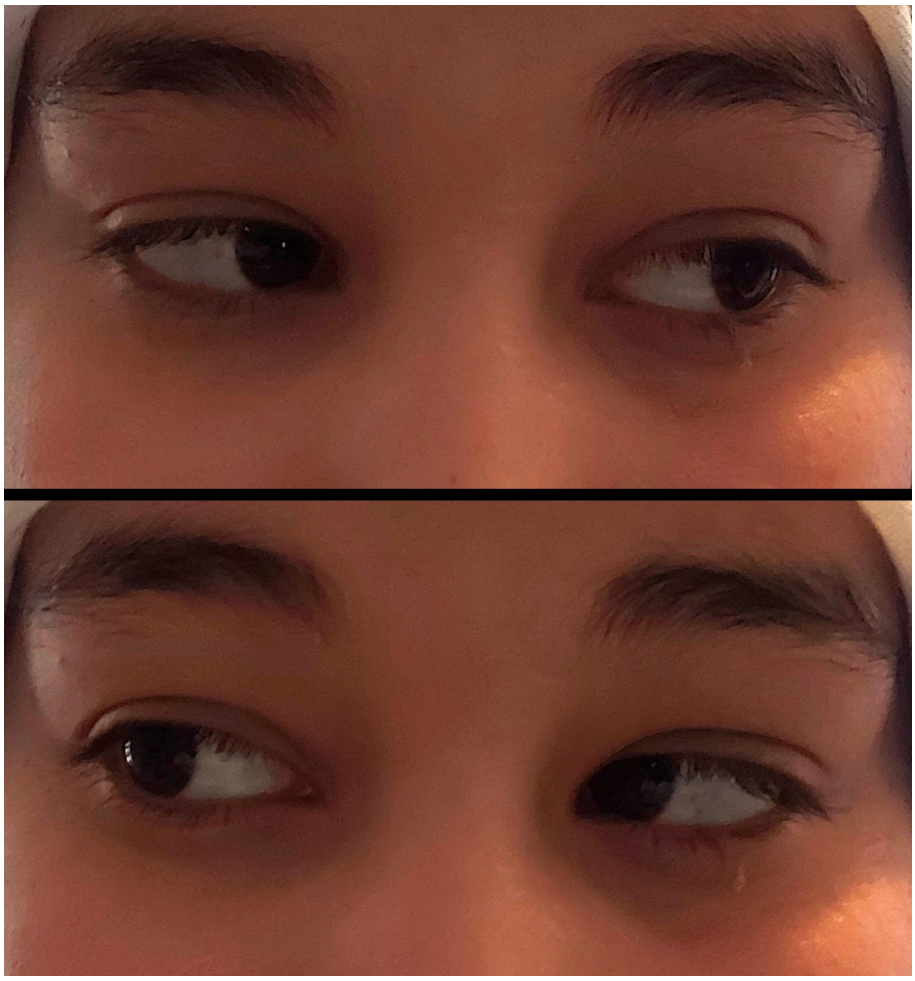

FIGURE 5. Complete recovery of ocular motility after 1 month

emerges in bones but could also manifest within soft tissues. It is ranked as the second most prevalent bone cancer in the paediatric population [1]. Management of this rare disease involves a multidisciplinary approach, including surgical resection and radiotherapy, and may as well be associated with chemotherapy [2].

Skull bones metastatic Ewing's sarcoma is seen frequently, but primary Ewing rarely involves the cranium [3]. This specific location accounts for only 1% of all cases. It affects predominantly the temporal bone, with the parietal bone and occipital bone being the subsequent most frequently involved sites [4].

Only a few cases of occipital bone involvement in primary Ewing sarcoma have been reported in the literature. In contrast to our patient, who exhibited only swelling of the posterior cranium, all documented cases reported severe headaches. Management of Occipital Ewing sarcoma involves a combination of cytotoxic drugs and surgical reduction or radiotherapy, or both [1]. Cranioplasty after resection of the tumor can be performed using various techniques and materials, such as an allogenic or autologous bone graft or an endoprosthesis. Methyl Methacrylate (MMA) is commonly employed as a cranioplastic material due to its widespread availability and cost-effectiveness [5].

In all the series where MMA has been used, there were mainly no severe post-operative complications described. In 1985, Vangool conducted a descriptive study that included 45 patients who had undergone MMA cranioplasty. No complications were reported related to the prosthesis, such as infection or disturbed wound healing [6].

Counting the side effects of MMA described in the literature, one could only mention those stated by Azmi et al., where 26.5% of all the patients included presented with a headache after surgery, but 85% of the patients improved after two months of proper treatment. The remaining patients required regular medication. Only one patient of the series experienced an exacerbation of headaches accompanied by vomiting in the immediate post-operative period but fully recovered within one month [5].

To date, there have been no documented cases where MMA cranioplasty has resulted in a complication such as intracranial hypertension (IH), as it was described in our patient.

Considering the definition of idiopathic intracranial hypertension (IIH), our case meets the predefined criteria of the disease [7]. Thus, it cannot be classified as such, given the clinical context in which the symptoms occurred. Only conjectures may be put forward as a possible remedy to the pathophysiological mechanism behind our patient's condition, which could be similar to that of IIH.

The pathophysiology of IIH is admittedly quite complex, invoking the restriction of venous drainage pathways, congestion of the glymphatic system, and blockage of the cerebrospinal fluid (CSF) lymphatic drainage pathway. CSF loaded with brain parenchyma waste would be eliminated in part via lymphatic vessels present in the dura mater [8]. The same pathophysiological model can be used to explain the clinic of our case.

Hence, it is plausible that the heat produced through the auto-polymerization of methyl methacrylate during its preparation could induce thermal damage to the underlying brain, given its exothermic reaction [5]. This could potentially result in congestion of the underlying venous circulation. The origin of this hypertension could be attributed to the waste product of MMA circulating in the cerebrospinal fluid, which is eliminated through the lymphatic vessels described earlier.

The symptoms in our patient exclusively emerged after the surgical intervention, aligning with those characteristics of IIH. This latter typically exhibits signs such as papilledema, headache, pulsatile tinnitus, and transient visual obscuration. In 20% of cases, patients may also present with horizontal binocular diplopia in cases of abducens nerve (VI) paralysis [7].

The VI cranial nerve (CN) palsy has previously been described in IIH among children. Findings of an American study in 2015 suggest an incidence of CN VI palsy in paediatric IH patients of approximately 12%. Furthermore, papilledema was an associated finding, which resolved ultimately after treatment [9]. Observations that were concurrent with our case.

CONCLUSION

This unique case challenges existing literature, as intracranial hypertension after MMA cranioplasty has not been previously documented. The pathophysiological mechanism is hypothesized to involve thermal damage from the exothermic reaction during MMA preparation, leading to venous congestion and interference with cerebrospinal fluid drainage. The manifestation of symptoms aligns with idiopathic intracranial hyperten-

sion (IIH) characteristics, emphasizing the need for careful consideration of complications associated with cranioplasty materials. This case contributes to our understanding of rare postoperative complications and underscores the importance of tailored treatment approaches for optimal patient outcomes.

Conflicts of interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Author contributions

T.A: patient care and follow-up, bibliographic research, and writing of the manuscript; S.S.: bibliographic research, and writing of the manuscript; M.Y.: bibliographic research, supervision and correction; O.A.: bibliographic research, supervision and correction.

Acknowledgments

None.

Funding

None declared.

REFERENCES

1. Riggi N, Suvà ML, Stamenkovic I. Ewing's Sarcoma. N Engl J Med. 2021; 384(2): 154–164, doi: 10.1056/NEJMra2028910, indexed in Pubmed: 33497548.
2. Grünewald TGP, Cidre-Aranaz F, Surdez D, et al. Ewing sarcoma. Nat Rev Dis Primers 2018, 4(1):5. 2018; 4(1): 5, doi: 10.1038/s41572-018-0003-x, indexed in Pubmed: 29977059.
3. Garg A, Ahmad FU, Suri A, et al. Primary Ewing's sarcoma of the occipital bone presenting as hydrocephalus and blindness. Pediatr Neurosurg. 2007; 43(2): 170–173, doi: 10.1159/000098397, indexed in Pubmed: 17337936.
4. Singh GR, Choudhary V, Agrawal R. Primary Ewing Sarcoma of Sphenoid Bone with Intracranial Extension: A Common Tumour at an Uncommon Location. J Clin Diagn Res. 2017; 11(2): ED15–ED16, doi: 10.7860/JCDR/2017/23541.9275, indexed in Pubmed: 28384876.
5. Azmi A, Latiff AZ, Johari A. Methyl methacrylate cranioplasty. Med J Malaysia 2004. 2004; 59(3): 418–421, indexed in Pubmed: 15727391.
6. van Go. Preformed polymethylmethacrylate cranioplasties: report of 45 cases. J Maxillofac Surg. 1985; 13(1): 2–8, indexed in Pubmed: 3856619.
7. Robelin F, Lenfant M, Ricolfi F, et al. Idiopathic intracranial hypertension: From physiopathological mechanisms to therapeutic decision]. Rev Med Interne 2022. 2022; 43(11): 661–668, doi: 10.1016/j.revmed.2022.08.012, indexed in Pubmed: 36114039.
8. Donnet A. Physiopathologie de l'hypertension intracranienne idiopathique. Revue Neurologique. 2021; 177: S150–S151, doi: 10.1016/j.neurol.2021.02.056.
9. Reid JE, Reem RE, Aylward SC, et al. Sixth Nerve Palsy in Paediatric Intracranial Hypertension. Neuroophthalmology. 2016; 40(1): 23–27, doi: 10.3109/01658107.2015.1117498, indexed in Pubmed: 27928378.