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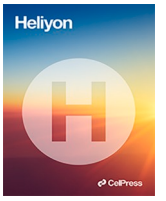

Case report

Sphenoidal pneumosinus dilatans associated compressive optic neuropathy: A case series of four adolescent patients

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ABSTRACT

Visual impairment caused by pneumosinus dilatans (PSD) among adolescents is a rare condition, which is associated with a high blinding rate due to the lack of clinical manifestations and effective treatment. The use of magnetic resonance imaging (MRI) and computed tomography (CT) may be helpful in diagnosis of PSD, and the endoscopic transnasal optic nerve decompression (ETOND) can improve the vision of PSD patients with visual impairments. This case series report detailing the diagnosis and treatment of visual impairments caused by PSD has improved clinicians' understanding of this disease and helped reduce misdiagnoses and missed diagnoses. This article also highlighted the efficacy of this less invasive technique in managing optic nerve compression caused by sphenoidal pneumosinus dilatans (SPSD) in pediatric patients. In addition, this report proposed a novel aspect to explore the etiology of PSD and suggested the significance of proper terminology use when describing different PSD conditions, as well as possible future mechanisms explorations.

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1. Introduction

Pneumosinus dilatans (PSD) is a rare condition characterized by the pathological hyperaeration and expansion of the paranasal sinuses, and the etiology of which remains obscure. The condition was first described in 1898 by Meyes [1]. In 1918, Benjamins proposed the term "pneumosinus dilatans" [2]. As of 2023, fewer than 150 cases have been reported in English literature. Moreover, adolescent cases are very scarce since PSD is more prevalent in male adults aged 20–40 years [3]. PSD has been reported that may lead to function disorders [4], while vision loss is the most serious one [5]. Endoscopic transnasal optic nerve decompression (ETOND), a minimally invasive optic nerve decompression technique [6], may be a potential way to treat PSD patients with visual impairments. However, there has been no study assess the effectiveness of this surgery on the treatment of PSD patients with seriously function disorders. Therefore, this cases series reported 4 cases of adolescent PSD patients with visual impairments who underwent ETOND.

2. Results

2.1. Patients' characteristics

Four adolescents with sphenoidal pneumosinus dilatans (SPSD) developed visual impairments were identified. The mean age of cases was 12.75 ± 2.06 years old (range from 11 to 15), and they were all male. Among 4 cases, no medical history was reported, but one case reported a family history of SPSD. The most typical symptoms of all cases were visual impairments with no reasons. All cases were diagnosed by computed tomography (CT) and magnetic resonance imaging (MRI) (Table 1).

2.2. Cases report

Case 1 was an 11-year-old Asian boy who presented with a blurred vision of the left eye for six months. No history of substance use, smoking, or drinking were reported. The initial diagnosis at local clinics concluded optic nerve atrophy and optic neuritis of the left eye. Visual acuity was 1.0 logMAR (logarithm of the minimum angle of resolution) in both eyes at the initial visit, as measured by Logarithmic Visual Acuity Chart. Symptoms persisted after three days of intravenous injection (IV) of 500 mg methylprednisolone sodium succinate. In the last ten days, the rapid decrease in visual acuity exacerbated the loss of light perception in the left eye before being admitted to our hospital. Before surgery, physical examinations diagnosed blindness in the left eye, while vision in the right eye was normal. CT and MRI revealed hyperaerated sphenoid sinuses, optic canal narrowing of both sides and left sinusitis (Fig. 1a and b). ETOND was performed 9 days after hospital admission.

Under general anesthesia, cotton pledgets soaked in 3 ml epinephrine (concentration of 1/1000) and 30 ml saline were placed in all nasal cavities every 5 minutes three times before surgery. The procedure began with a nasal endoscopy to assess the nasal anatomy and identify the path to the sphenoid sinus. Following identification, the sphenoid sinus was accessed via a transnasal approach. Ethmoidectomy was performed to expose the lamina papyracea. After performing sphenoidotomy, hyperaerated sphenoid sinus was observed, and the optic canal was suspended in the sphenoid sinus (Fig. 2a). The optic canal narrowing site was at the intracranial end, as revealed by preoperative radiology results. The optical nerve suspended in the bones adjacent to the superior and inferior of the optic canal was thinned using a 4 mm diamond drill at 40,000 rpm (Fig. 2b). The optic canal was widened 180° to decompress the pressure. The operative site was packed with hemostatic material along with antibiotics.

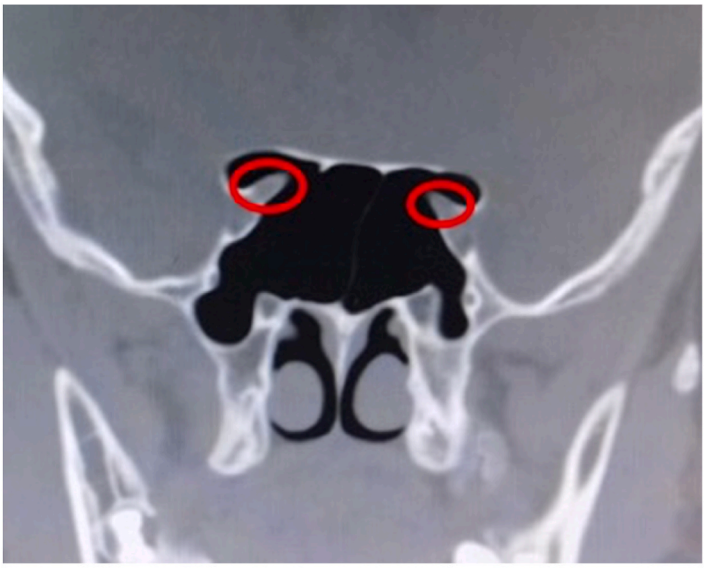
Slight light perception at the nasal visual field appeared one day after surgery. Three days after surgery, visual function was assessed using the "counting fingers" (CF) ordinal scale, which is suitable for patients with severe visual impairments who are unable to recognize characters on a standard eye chart [7], and the visual acuity was restored to CF/40 cm. Visual acuity reached CF/1 m seven days after surgery. Left eye visual acuity improved to 0.03 logMAR and 0.04 logMAR thirty days and forty days after surgery, respectively. Follow-up MRI confirmed the absence of optic nerve compression and normal left optic canal. Follow-up visits for 10 months observed sustained normal visual function.

Case 2 was a 14-year-old Asian boy who complained about painless blurred vision in both eyes over 2 months. Six days after the appearance of optical symptoms, the patient experienced sudden visual loss for 24 hours and was admitted to the local hospital. The patient underwent glaucoma surgery for both eyes at the local hospital. Later after surgery, the patient was diagnosed with retrobulbar neuritis, and was administered 500 mg methylprednisolone (IV) for three days. Visual acuity improved after treatments. However, symptoms relapsed when treatments ceased. The patient visited the ophthalmology clinic, where he received 500 mg intravenous

Table 1
Patients characteristics.

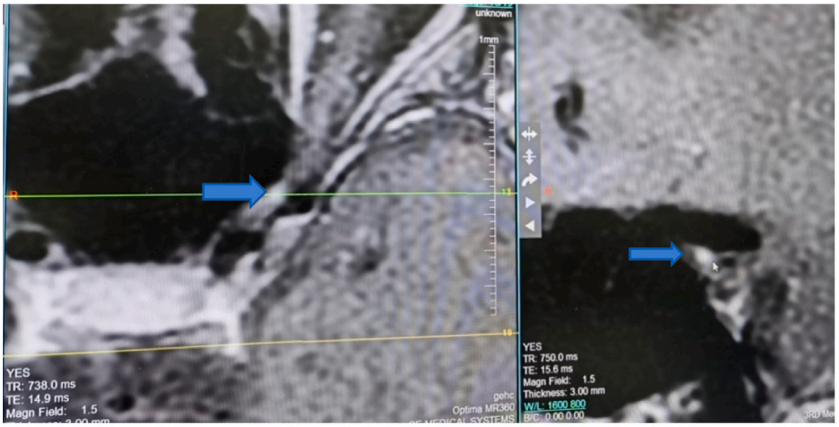
	Sex	Age	Race	Medical History	Family History of SPSD	Symptoms	Duration of symptoms	Diagnose method
1	Male	11	Asian	None	No	1. Blurred vision of the left eye 2.Decrease in visual acuity 3.Loss of light perception in the left eye	Six month	CT and MRI
2	Male	14	Asian	None	No	1.Painless blurred vision in both eyes 2.Decrease in visual acuity	Over 2 months	CT
3	Male	11	Asian	None	Yes	Painless decrease in visual acuity	One month	CT
4	Male	15	Asian	None	No	Blurred vision	48 months	CT and MRI

a: Computed tomography scan of Case1:Optic canal narrowing of both sides



label:Optic canal

b: Magnetic resonance imaging of Case 1: Optic canal narrowing of left side



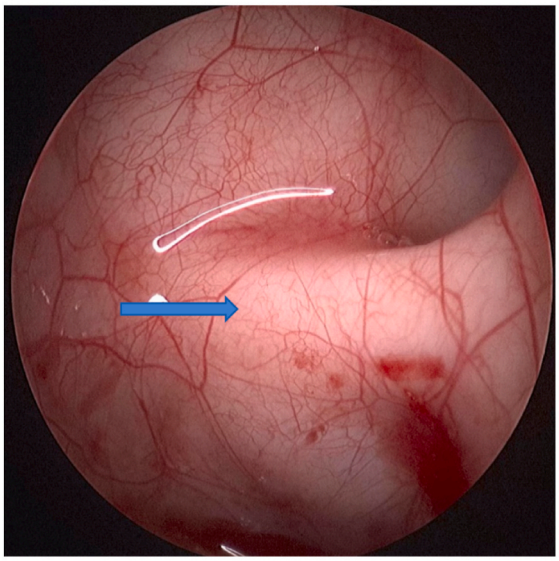
label: optic canal

Figure 1. The images of CT and MRI of *case 1*. Circled in red shows optic canal narrowing of both sides; the blue arrow indicates optic canal narrowing of left side.

methylprednisolone sodium succinate for four days with significant visual improvement. One month after the second hospital visit, the patient presented to our hospital with elevated intraocular pressure. He was administered 250 mg methylprednisolone sodium succinate (IV) for ten days before surgery. CT scan illustrated hyperaerated sphenoid sinuses (Fig. 3). The uncorrected visual acuity of the right eye was 0.04 logMAR and that corrected visual acuity was 0.1 logMAR, respectively. Similarly, visual function of the left eye was assessed using the CF ordinal scale. Visual acuity of the left eye was CF/5 cm, with no significant improvement after correction. Based on his physical examination results, he was diagnosed with bilateral compressive optic neuropathy due to SPSD. The operation was conducted one month after being admitted to our hospital.

Similar procedures of preoperative preparation, endoscopic identification, ethmoidectomy and sphenoidotomy to *case 1* were performed. During ETOND, dilated sphenoid sinuses were observed along with suspended optic nerves, and the optic canal was widened 180° (Fig. 4a and b). The optic nerve narrowing site was decompressed using a 4 mm diamond drill followed by 2 mm drill for polishing. Postoperative care was similar to case 1. Visual acuity was 0.1 logMAR (left eye), and 0.05 logMAR (right eye) one day after surgery. Six months after surgery, visual acuity remained the same. Postoperative pathological analysis concluded mild bone tissue degeneration. Additionally, genetic tests ruled out possible mitochondrial retinal diseases.

a Endoscopic identification of the left optic canal



b Endoscopic Transnasal Optic Nerve Decompression: left optic canal was widened 180°

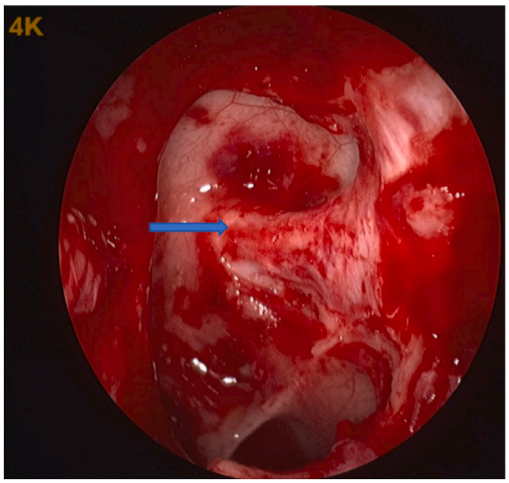


Figure 2. Endoscopic Transnasal Optic Nerve Decompression of *case 1*. a: endoscopic identification of the left optic canal; b: left optic canal was widened 180°.

Case3 was a 11-year-old Asian boy who presented to our hospital with complaints about a painless decrease in visual acuity for one month. A family history of SPSD was reported by the patient, whose father was diagnosed with hyperaerated sphenoid sinuses. Initial symptom of decreased right eye visual acuity was discovered during the routine physical examination at school. Right eye visual acuity was CF/10 cm that measured using CF ordinal scale. The patient underwent fundus photography, visual tests, and MRI during the first visit to the local hospital. The initial physical examinations disclosed optic nerve degeneration, optic nerve thinning, and optic nerve sheath widening of the right eye. Visual evoked potential (VEP) results showed decreased wave amplitude of the right eye. The patient was referred to our hospital one month after the appearance of the ocular symptoms. CT scan at our hospital demonstrated optic nerve atrophy of the right eye and right anterior clinoid process pneumatization. Visual acuity of the left eye was normal, while the right eye was CF/10 cm. The patient was diagnosed with SPSD and compressive optic neuropathy. The patient's condition necessitates surgical decompression. Therefore, ETOND was performed to decompress the lateral optic nerve and the orbital wall. The similar surgical procedure to *case 1* was conducted. Intraoperatively, right optic nerve canal suspension was observed. The optic canal was widened 180° to decompress the pressure. The patient's right visual acuity improved to CF/30 cm one day after surgery. Visual acuity of the left eye remained normal. Nine days after surgery, the visual acuity remained stable.

Case 4 was a 15-year-old Asian boy who suffered from blurred vision for two years. Two months before being referred to our hospital, the patient was diagnosed with bilateral optic neuritis and optic atrophy by the local hospital. There was no significant symptom amelioration after non-invasive treatment at the local hospital. The patient underwent visual tests, a CT scan, and MRI after

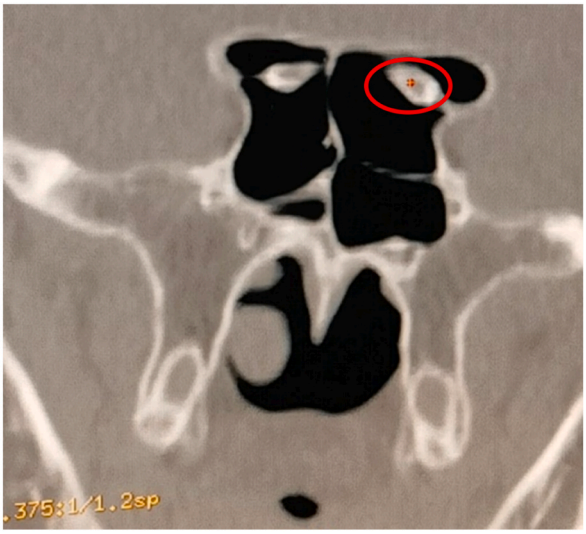


Figure 3. The computed tomography scan of *case 2*. Circled in red shows that the optic canal was suspended in the sphenoid sinus.

being admitted to our hospital. Upon admission, CF ordinal scale examination showed the visual acuity of his right eye was CF/5 cm. His left eye showed better acuity at 0.4 logMAR. CT scan disclosed enlargement and pneumatization of the anterior clinoid process, with more severe symptoms on the right side than on the left side. MRI results concluded bilateral sphenoidal pneumatization and fluid in the optic nerve sheath. The patient's preoperative visual acuity decreased to 0.01 logMAR in the left eye. His right eye is only able to see hand movement.

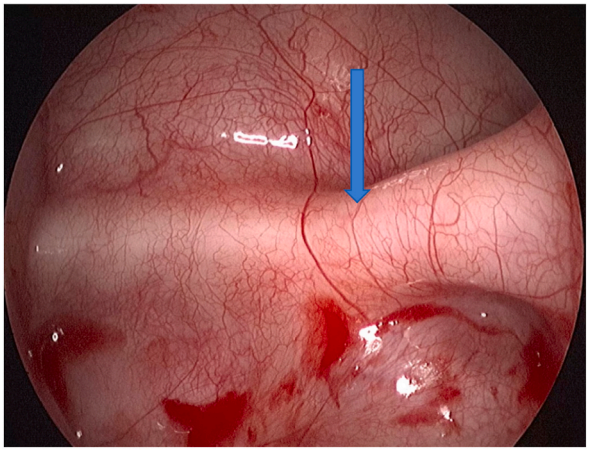
The diagnosis of SPSD and compressive optic neuropathy was made with recommendations for decompression surgery. Under general anesthesia, the patient underwent ETOND, during which the right optic nerve canal was identified, and thinning optic nerve due to compression was observed. A wider exposure (270°) of the optic canal was made using a diamond drill (Fig. 5). Intraoperative monitoring ensured the preservation of visual function throughout the procedure. Three days after surgery, the patient's uncorrected right eye visual acuity was CF/20 cm, and the corrected visual acuity was 0.01 logMAR. Left eye corrected acuity was 0.05 logMAR, with accurate light perception in both eyes. Two weeks postoperative, left visual acuity turned to be 0.08 logMAR, while the right visual acuity remained the same. Three-month follow-up visit showed stable visual function.

3. Discussion

This report presents four rare cases of adolescents with hyperaeration of the sphenoid sinus. Although some speculations have been proposed, the etiology and pathogenesis of PSD remain obscure and may be multifactorial. The most accepted and evidenced theory is the ball valve theory, involving the one-way valve mechanism [8]. Briefly, the ostial polyp or other mucosal abnormalities lead to the obstruction of the sinus ostium and elevated sinus pressure. Long-term pressure gradient eventually resulted in sinus expansion. Other theories with less supported evidence are fibro-osseous dysregulation [9], mucocele drainage [10], tramas [11], meningioma [12], and vitamin D deficiency [13]. The occurrence of PSD is also associated with other complications, such as port wine stain [14], arachnoid cysts, Melnick-Needles syndrome, Klippel-Trenaunay-Weber syndrome, and Sturge-Weber syndrome [15]. Earlier proposals regarding gas-forming bacteria have failed to evident the speculations and have therefore been discounted. In our cases, all patients presented with SPSD are adolescents, leading us to consider the theory of hormonal change during puberty, which is, however, not supported by current research [16]. It is also rational to speculate that the disease onset in adolescence is because the sphenoid sinus is not fully developed until puberty [17]. Furthermore, our *case 3* reported his father was also diagnosed with SPSD, suggesting the potential inherited nature of the disease. As most research demonstrated, males are more affected by PSD. Future research may also attempt to identify possible associated genetic factors of the Y chromosome. Our cases shed more light on the potential causes of the disease. Nevertheless, research in exploring the possible mechanisms is highly warranted.

The diagnosis of PSD also remains challenging due to the rarity of the disease. As of 2020, previous literature reviews have identified 171 cases of PSD since the first diagnosis [15,16,18]. Since 2020, eleven more cases with varying ages and causes have been reported globally in literature [19–29]. Only two cases have been diagnosed thus far in China [21,29]. Most reported PSD cases are males aged 20–40 years old. The expanded frontal sinus is the most common condition in PSD cases (63 %), followed by the sphenoid sinus (25 %), maxillary sinus (19 %), and ethmoid sinus (18 %) [16]. On the other hand, patients may not seek medical treatment since most PSD are benign and asymptomatic. Patients may not notice the symptoms as well. The most frequently reported symptoms are swelling of the forehead, decreased vision, and headache [15]. Therefore, the true incidence may be much higher than the currently reported rate. In addition, misdiagnosis is common as many health providers are unaware of the disease or familiar with the evaluation

a: Endoscopic identification of the left optic canal



b: Endoscopic Transnasal Optic Nerve Decompression: left optic canal was widened 180°

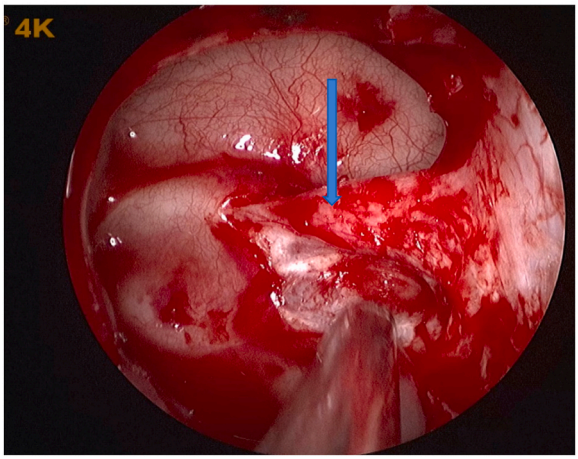


Figure 4. Endoscopic Transnasal Optic Nerve Decompression of *case 2*. a: endoscopic identification of the left optic canal; b: left optic canal was widened 180°.

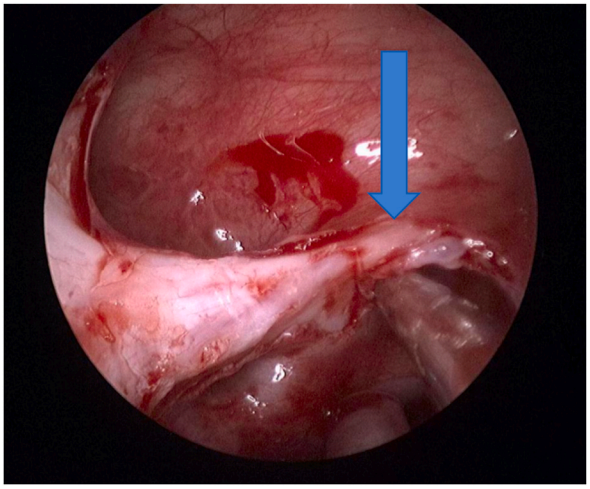


Figure 5. Endoscopic Transnasal Optic Nerve Decompression of *case 4*. The blue arrow indicates left optic canal was widened 270°.

approach.

Although the underlying mechanism and prevalence are understudied, the treatment strategy has improved substantially in recent decades. Intravenous corticoid administration is a conservative approach to managing optic neuropathy when surgical options are not available or not recommended [30]. When surgery is advised, ETOND and craniotomy are the most commonly performed surgical procedures to decompress the optic nerve. The prior approach is preferred over craniotomy in several circumstances for better prognosis and aesthetic outcomes [31]. Adolescence is a critical period for physiologic and psychological development. Invasive treatment for PSD may impact adolescents' physical and mental health due to the potential damage and facial scars. Unlike open procedures that may lead to brain retraction and scare formation, the minimally invasive nature of ETOND, along with its favorable aesthetic outcomes, has emerged as a valuable option for adolescent patients with optic nerve compression caused by SPSD. We reported four cases of SPSD with successful ETOND and significant symptom resolution, advocating the application of such a procedure in adolescent SPSD patients. Since optic neuropathy is potentially irreversible, we recommend early ETOND in adolescents when visual decline is reported, and radiology diagnosis shows an enlarged sphenoidal sinus.

More precise definitions and terminology are critical in diagnosis and revealing the potential mechanism. PSD are mainly aesthetic concerns, while SPSD is commonly associated with functional impairments. Paranasal sinuses consisted of frontal, ethmoid, sphenoid, and maxillary sinuses [32]. Unlike other sinuses, hyperaeration in the sphenoid sinus may result in visual impairment due to its proximity to the optic nerve and carotid artery [33]. Previous reviews indicated that more than half of the patient with SPSD lost their vision [3]. All four cases reported in this article suffered from visual impairment, the symptoms of which varied from decreased visual acuity to blindness. Endoscopic results displayed suspended optic nerves. The rapid and sudden visual declines, some to the degree of blindness, highlight the distinct nature of SPSD. On the other hand, the sphenoid sinus is the most posterior paranasal sinus, and the complexity of surgical management is also higher. Using PSD to describe all hyperaerated sinuses analogously may be too general and vague. Thus, we propose a more accurate term, "Sphenoidal Pneumosinus Dilatans," to describe hyperaerated sphenoid sinus. The more targeted terminology may pave the way for future classification, diagnosis, as well as theoretical explorations.

4. Limitations

There were some limitations in the study. First, the case sample was small which may cause bias in results. Secondary, the study was a retrospective observational study and due to the limitation of this study design, the findings only may infer association rather than causation. However, SPSD with visual impairments is a seriously rare disease, and few studies explored the diagnose method and proper treatment. Our findings can provide some evidence to clinical diagnose and define the disease, and also strongly recommend the ETOND as the primary treatment to improve the symptoms for SPSD patients. In addition, examinations were not performed comprehensively in this research, limiting the validation of certain etiological speculation. Future study should include investigations, such as blood tests, to validate potential mechanisms.

5. Conclusion

We reported four adolescents diagnosed with SPSD. ETOND was successfully performed to treat compressive optic neuropathy. All cases reported an increase in postoperative visual acuity. Also, we proposed the term "Sphenoidal Pneumosinus Dilatans," to describe hyperaerated sphenoid sinus. ETOND is highly recommended for adolescent PSD patients due to its minimally invasive nature. Proper diagnosis of SPSD and fast surgical decisions are required before severe visual function impairments occur.

Ethics statement

This is a single center retrospective observational case series study conducted in the Chinese People's Liberation Army General Hospital and Medical School. Adolescents diagnosed with SPSD were recruited from December 2022 to December 2023. Informed consent was obtained from all patients or their guardians. Patients' demographic characteristics and medical history were collected in the medical record. Endoscopic Transnasal Optic Nerve Decompression was performed in all patients. The study was approved by the Ethics Committee of Chinese PLA General Hospital (S2018-088–01).

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None.

Data availability statement

The data are available from the corresponding author on reasonable request.

CRediT authorship contribution statement

Xuejun Zhou: Writing – review & editing, Writing – original draft, Investigation, Data curation. **Quangang Xu:** Data curation. **Buhuan Zhang:** Methodology. **Yongzhe Liu:** Validation. **Xinying Liu:** Visualization. **Songfeng Wang:** Formal analysis. **ShiMing Yang:** Writing – review & editing, Validation, Supervision, Resources. **Xiaolu Wang:** Writing – review & editing, Writing – original

draft, Visualization, Validation, Resources, Methodology, Investigation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix ASupplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2024.e38763.

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