## **BRIEF COMMUNICATION**

# Unusual Posterior Hyaloid Strand in a Young Child with Optic Disc Pit Maculopathy: Intraoperative and Histopathological Findings

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#### Abstract

**Background:** The mechanism responsible for optic disc pit maculopathy is unclear, but abnormal vitreous structures, including the anomalous Cloquet's canal at the optic disc pit, have been suggested as important factors.

**Case:** We report the intraoperative and ultrastructual findings of an unusual posterior vitreous strand in the eye of an 8-year-old girl with optic disc pit maculopathy.

**Observations:** The patient presented with decreased vision in the left eye. Examination of the left eye revealed a best-corrected visual acuity (VA) of 0.08 and a macular detachment associated with an optic disc pit. Vitrectomy was performed with the adjunctive use of triamcinolone acetonide intraoperatively. The presence of an unusual posterior hyaloid strand tightly attached to the margin of the optic disc pit was noted. An unusual movement of this strand was observed during the surgery. The strand was excised, and fluid–gas exchange was performed using gas tamponade with 20% SF<sub>6</sub>. After 12 months, a complete macular reattachment was obtained, with the VA improving to 1.2. Electron microscopic examination of the removed strand revealed abundant thick collagen fibrils with a frame of fine fibrils.

**Conclusion:** The unusual posterior vitreous strand connected to the optic disc pit may have contributed to the pathogenesis of maculopathy in this young child. **Jpn J Ophthalmol** 2005;49:264–266 © Japanese Ophthalmological Society 2005

Key Words: Cloquet's canal, macular detachment, optic disc pit, triamcinolone acetonide, vitrectomy

### Introduction

Optical coherence tomography (OCT) has recently contributed greatly to the detection of the two-layered structure of optic disc pit maculopathy, consisting of a posterior retinoschisis and outer layer detachment.<sup>1</sup> However, the pathogenesis of optic disc pit maculopathy remains unclear. Some studies have implicated the vitreous and the condition of the posterior hyaloid as causes of the serous macular detachment associated with optic disc pits.<sup>2–5</sup> Hasegawa and colleagues,<sup>3</sup> using scanning laser ophthalmoscopy, observed a cystlike structure terminating at the pit in the premacular vitreous. In the present case, during vitrectomy in which triamcinolone acetonide (TA) had been used to highlight the posterior hyaloid membrane, we observed an unusual posterior hyaloid strand connected to the optic disc pit. We report a peculiar movement of this vitreous strand during surgery, and the ultrastructural features of the excised strand. This case has been included in a long-term study on the results of vitreous surgery for optic disc pit maculopathy (submitted for publication).

#### **Case Report**

An 8-year-old girl presented with decreased vision in the left eye after blunt trauma caused by a volleyball. Her best-

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corrected visual acuity (VA) in the left eye was 0.08. An optic disc pit was detected on the temporal side of the optic disc, associated with an oval-shaped, shallow macular detachment covering the area between the superior and inferior vessel arcade. The macular detachment appeared to communicate with the pit. A round, two-disc diameter, serous retinal detachment was observed in the center of the macula, with cystic changes. OCT (Zeiss-Humphrey, San Leandro, CA, USA) revealed a separation of the inner retinal layers that appeared to connect with the optic disc, as well as a defect in the outer layer at the macula and a detachment surrounding the hole.

Because of the progression of the macular detachment and the enlargement of the outer macular hole over the next 4 months, we decided to perform vitrectomy. Informed consent was obtained from the parents after an explanation of the procedures and prognosis. The use of TA intraoperatively enabled us to clearly visualize the posterior hyaloid membrane. During the induction of the posterior vitreous detachment (PVD), a hyaloid strand was detected tightly adhering to the margin of the disc pit (Fig. 1A). Part of the

tonide was used to highlight the posterior hyaloid. A During the induction of posterior hyaloid separation by suction with the vitreous cutter, the posterior hyaloid can be seen tightly attached to the edge of the optic disc pit. B An unusual posterior hyaloid strand can be seen after the separation of the posterior hyaloid over the posterior pole. C The strand was quickly sucked into the pit immediately after the sclerotomy site was closed by the forceps but was later retrieved with forceps for electron microscopic examination.

strand was left attached to the edge of the pit after the removal of the posterior hyaloid membrane over the posterior pole with a vitreous cutter (Fig. 1B). As the vitreous forceps was inserted through the sclerotomy site to grasp the strand at the pit, the strand was quickly sucked into the pit and almost disappeared (Fig. 1C). The strand was pulled out of the pit with the vitreous forceps and removed. Fluid-air exchange was performed followed by gas tamponade with 20%  $SF_6$ .

The patient's VA began to improve 1 month postoperatively, corresponding to the improvement in the retinoschisis and macular serous detachment. At 6 months postoperatively, the VA was 0.8. OCT performed at 9 months postoperatively showed resolution of the retinoschisis, but the serous detachment was still present. By 12 months, the VA was 1.2, and funduscopy and OCT showed a complete reattachment of the retina.

Electron microscopic examination of the excised specimen revealed abundant clusters of collagen fibers with a frame of fine fibrils (Fig. 2A). Most of the collagen fibrils, which appeared to be type I collagen, were 15 to 25 nm in



**Figure 2A,B.** Electron photomicrographs of the excised specimen. **A** Electron microscopic examination shows clusters of collagen fibers with a frame of fine fibrils. Very few cells are present. Bar =  $1.0 \mu m$ . **B** Under higher magnification, most of the collagen fibrils, which appear to be type I collagen, were 15 to 25 nm in diameter with a regular periodicity of 45–55 nm. Bar =  $0.1 \mu m$ .

diameter with a regular periodicity of 45–55 nm (Fig. 2B). Very few cells were present in the specimen.

### Discussion

Recent OCT findings have revealed that the macular detachment associated with an optic disc pit consists of a bilaminar structure. However, the mechanism responsible for the macular serous detachment remains unclear. Bonnet<sup>5</sup> reported that none of 25 eyes with macular serous detachment associated with an optic disc pit initially had a PVD, and two of the eyes had a spontaneous reattachment of the macula following the development of a PVD. Several investigators have discussed the efficacy of vitrectomy and gas tamponade to treat optic disc pit maculopathy.<sup>3-5</sup> In our studies, vitrectomy using modern surgical techniques to create a PVD in young patients and gas tamponade without

laser photocoagulation were successful in reattachment of the macula with an improvement in central vision in 10 out of 11 eyes with optic disc pit maculopathy. However, most of the eyes required almost 1 year of recovery to attain this state (submitted for publication). These observations support the concept that vitreous traction may have an important role in the development of macular detachment associated with optic disc pits.

Akiba and coauthors<sup>2</sup> reported that 11 out of 15 eyes with optic pit maculopathy without a PVD had an anomalous Cloquet's canal that was markedly condensed and terminated at the margin of the pit. During ocular movements, they observed a back-and-forth movement of the anomalous Cloquet's canal and a pulsating translucent membrane that covered the pit. Several case reports have described similar abnormal structures anterior to the pit, based on scanning laser ophthalmoscopic observations or intraoperative findings.<sup>3,4</sup>

The intraoperative use of TA to observe the posterior hyaloid appears to be a useful technique to detect abnormal adhesions of the vitreous to the margin of the disc pit, as in our case. The peculiar sucking of the strand into the pit during the insertion of the vitreous forceps into the eye suggests that the optic disc pit may connect to the subarachnoid space, resulting in suction from the subarachnoid space produced by an imbalance between the intraocular and the subarachnoid pressures.

Histological examination of the posterior hyaloid strand that was connected to the pit showed abundant clusters of thick collagen fibers with a frame of fine fibrils and very few cells. This feature of collagen is different from that found in the native vitreous collagen, and is consistent with the observations of previous reports<sup>2-4</sup> describing the presence of condensed membranes in anomalous Cloquet's canals attached to optic disc pits. However, we did not observe similar vitreous strands in most of the eyes with optic disc pit maculopathy in our clinical series.

Patient age at the onset of the retinal detachment associated with optic disc pits is variable, with a mean age of 30 years. Our present patient was exceptionally young, suggesting that unusual posterior vitreous strands connected to the optic disc pit might contribute to the pathogenesis of maculopathy in some patients with optic disk pit.

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