Case Report

Sheathotomy without Separation of Venule Overlying Arteriole at Occlusion Site in Uncommon Branch Retinal Vein Occlusion

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Two cases of uncommon branch retinal vein occlusion (BRVO) with vein overlying artery at occlusion site that can be found in less than 1 % who underwent retinal venule sheathotomy without separation of retinal vessel for decompression of BRVO were reported. Both patients had macular hemorrhage, edema, and area of macular capillary nonperfusion. Visual acuity (VA) worsened to 6/60 in both cases. The retinal venules were dissected around the crossing site without separation of retinal vessels. Intraoperative dilation, pulsation and restoration of downstream blood flow of the involved venules were observed. In the first patient, at 1 day, 2 weeks, and 6 weeks postoperatively, VA improved to 6/36, 6/9, and 6/6, respectively, and remained unchanged at 12 months postoperatively. In the second patient, VA improved to 6/24 on the first day post-operatively and improved to 6/18 at 1 week follow up and continuously improved to nearly normal at 2 months postoperatively then patient lost contact. Postoperative fundus fluorescein angiogram showed dilated and improved perfusion with decreased macular edema in both cases. Optical coherent tomography confirmed remarkable reduction of retinal thickness (from 874 microns preoperatively to 420 microns at 1 week postoperatively) in the second patient. Retinal venule sheathotomy without separation of retinal vessel for decompression of BRVO with venule overlying arteriole at occlusion site could be effective for improving VA and decreased macular edema.

Keywords: *Sheathotomy, Branch retinal vein occlusion, Venule overlying arteriole*

J Med Assoc Thai 2005; 88 (Suppl 9): S143-50
Full text. e-Journal: *http://www.medassocthai.org/journal*

Branch retinal vein occlusion (BRVO) is the second most common vascular disorder of the retina and usually causes visual loss due to macular edema, macular ischemia and intraretinal hemorrhage. The occlusion sites in BRVO occurs typically at the arterio-venous (A/V) crossing site where the artery and vein are in close contact and share a common adventitial sheath. Previous studies showed that 0% to 1% of the crossing at the occlusion sites, the vein was located anterior to the artery[1-4].

The Branch Vein Occlusion Study(BVOS)[5-7], a randomized controlled trial, recommended grid argon laser photocoagulation for BRVO with macular edema with a visual acuity (VA) 20/40 or worse after follow up for 3 months because 65% of the treated group gained $\geq$ 2 lines of vision versus 37% of the control group. However, despite the visual improvement being statistically significant, the mean number of lines of vision gained at the 3rd year visit was 1.33 in the treated group and 0.23 in the control group. This may reflect a poor prognosis for patients with poor initial VA. More over, BVOS excluded patients with poor initial VA < 20/200 and patients with distinct area of macular capillary nonperfusion from the treatment[5-7]. The laser treatment may be effective for treating macular edema but may be ineffective for restoring blood flow in this group of patients. A new therapeutic approach, "arteriovenous adventitial sheathotomy (AAS)", is based on the fact that the occlusion is at the A/V crossing site. In 1988,

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Osterloh and Charles first performed vitrectomy with dissection of the adventitial sheath and separated overlying artery from a vein as an alternative approach that could restore venous blood flow, improving retinal perfusion and resolving intraretinal hemorrhage and edema[8]. Until 11 years later, more studies about AAS have been published with different study designs and modified technical details. Promising initial results and also complications have been reported with unclear exact mechanism[8-29].

This is a report of 2 uncommon BRVO cases with venule overlying arteriole at the obstructed A/V crossing site that underwent retinal venule sheathotomy without separation of retinal vessel for decompression of BRVO.

Case Report

Case 1

A 42-year-old man, with no underlying diseases, presented for a recent visual impairment of the left eye for 2 weeks. His VA was 6/6 in the right eye and 6/24 in the left eye on a snellen chart. On eye examination, anterior segment and intraocular pressure was unremarkable. Examination of the left fundus revealed a superotemporal BRVO with intraretinal hemorrhage, cotton wool spot and macular edema (Fig 1a). Fluorescein angiography (FA) revealed the location of the occlusion with a vein overlying the artery at A/V crossing site, macular non perfusion and macular edema (Fig. 1b). On 2 weeks consecutive follow up, VA of the left eye decreased to 6/60. After informed consent was

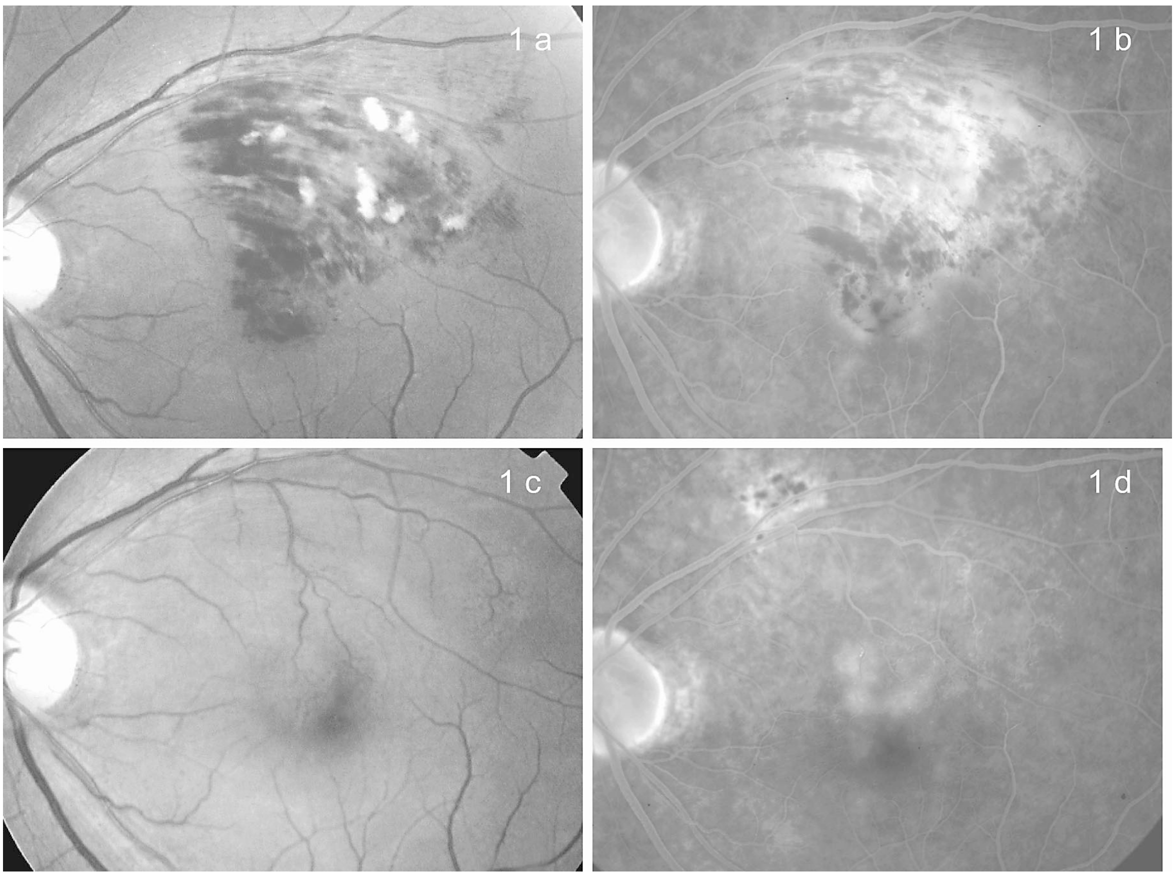

Fig. 1 Demonstrates fundus photographs and late phase fluorescein angiography of the left eye of case 1
1a) Preoperative fundus photograph shows occlusion of superotemporal branch of retinal vein with cotton wool spots, diffuse hemorrhages and macular edema
1b) Preoperative fluorescein angiography shows macular edema and non perfusion area
1c) Fundus photograph 11 months after surgery shows resolution of hemorrhages and macular edema
1d) Fluorescein angiography 11 months after surgery confirms findings of clinical examination, improvement of retinal perfusion and decreased macular edema with hypo- and hyperfluorescence area from retinal pigment epithelial alteration peripheral to occlusion site

obtained, the patient underwent sheathotomy at 4 weeks after the onset of visual symptoms. The procedure consisted of three port pars plana vitrectomy without posterior hyaloid removal because posterior vitreous detachment was present. A microvitreoretinal blade was used to dissect the retina and sheath around the overlying retinal vein and a bent microvitreoretinal blade was used to lift the retinal vein free from the retinal surface proximal and distal to A/V crossing site. Intraoperative dilation, pulsation and restoration of downstream blood flow of the involved venule were observed. Because of a strong adhesion between the vein and artery at the A/V crossing site, the surgeon judged that more aggressive maneuvers would result in laceration of retinal vessel. In this case the retinal vein was not separated from the artery. VA improved to 6/36 on the first day, 6/9 at 2 weeks and 6/6 at 6 weeks, postoperatively. After a 12 month follow-up, visual acuity was still 6/6. The postoperative fundus examination showed retinal vein dilatation and improvement of retinal perfusion with decreased macular edema (Fig. 1c), FA showed retinal vein dilatation, improvement of retinal perfusion with decreased macular edema and hyperfluorescence area from retinal pigment epithelial alteration peripheral to the occlusion site (Fig. 1d).

Case 2

A 45-year-old man, with a history of systemic hypertension and dyslipidemia, presented with progression of visual impairment of the left eye for 3 weeks. His VA was 6/6 in the right eye and 6/60 in the left eye on a snellen chart. On eye examination,anterior segment and intraocular pressure was unremarkable. Retinal examination revealed an inferotemporal BRVO with collateral vessel around the crossing site. (Fig. 2a) On FA, macular non perfusion and macular edema were also noted (Fig. 2b). Optical coherent tomography showed macular edema, the foveal retinal thickness was 874 microns (Fig 2c). After informed consent was obtained, the patient underwent sheathotomy 4 weeks after the onset of visual symptoms. The procedure was similar to the first case except the surgeon did not attempt to separate the vein from the artery at the A/V crossing site. Intraoperative dilation, pulsation and restoration of downstream blood flow of the involved venule were observed. During sheathotomy, a small hemorrhage from a small collateral vessel near the occlusion site was stopped by increased intraocular pressure. At 1 day and 1 week postoperatively, VA improved to 6/24, 6/18, respectively. At 1 week follow-up, the fundus examination revealed increased vein dilatation with decreased hemorrhage, edema and collateral vessel around the crossing site. (Fig. 2d) By the last phone contact with the patient at 2 months postoperatively before the patient was lost the follow up, visual acuity was continuously improved to nearly normal compared with the other eye. The postoperative FA at 1 week showed retinal vein dilatation and improvement of retinal perfusion with decreased macular edema and collateral vessel (Fig 2e). Postoperative OCT at 1 week revealed decreased foveal retinal thickness to 420 microns (Fig. 2f).

Discussion

It has been shown that almost all occlusions in BRVO occurred at the arteriovenous (A/V) crossing sites where the artery is positioned anterior to the vein. In 463 BRVO cases from 4 A/V crossing pattern studies, only 1 case had the vein anterior to the artery at occlusion sites (1% in 1 study[1] and 0% in 3 studies[2-4] but vein anterior to the artery at non occlusion A/V crossing sites occured in 30-40%[1-4].

The histopathologic study at the A/V crossing sites showed that artery and vein share a common wall, vein diameter was reduced as it passed the crossing site and patent venous lumen with a fresh or recanalized thrombus[30]. Although the exact mechanism by which venous occlusion occurs at pathologic A/V crossing is unclear, in the recent literature the pathogenic mechanisms of BRVO are postulated to consist of compression of the vein by the sclerotic artery. The abnormal flow downstream from the constriction may induce downstream turbulence, endothelial damage and secondary thrombosis in vein. The persistence of the constriction may lead to continued elevation of upstream venous pressure, edematous retinal changes and hemorrhage[4,31]. This may also occur in the vein cross artery at the occlusion site.

In the presented patients, at the occlusion site, a secondary branch vein was involved in the first case and a primary branch vein was involved in the second case. In the second case, visual loss was more rapid progressively, the fluorescein angiogram also showed a larger area of nonperfusion and large collateral branch around the crossing site near the optic disc. The clinical course after the onset of BRVO can be determined by the location of the BRVO in relation to the fovea, the extent of the involved venous drainage area, and the collateral drainage capacity from the area with compromised venous drainage to the adjacent areas of intact venous drainage[4]. Collateral maturation occurs over a period of 6 to 24 months after the

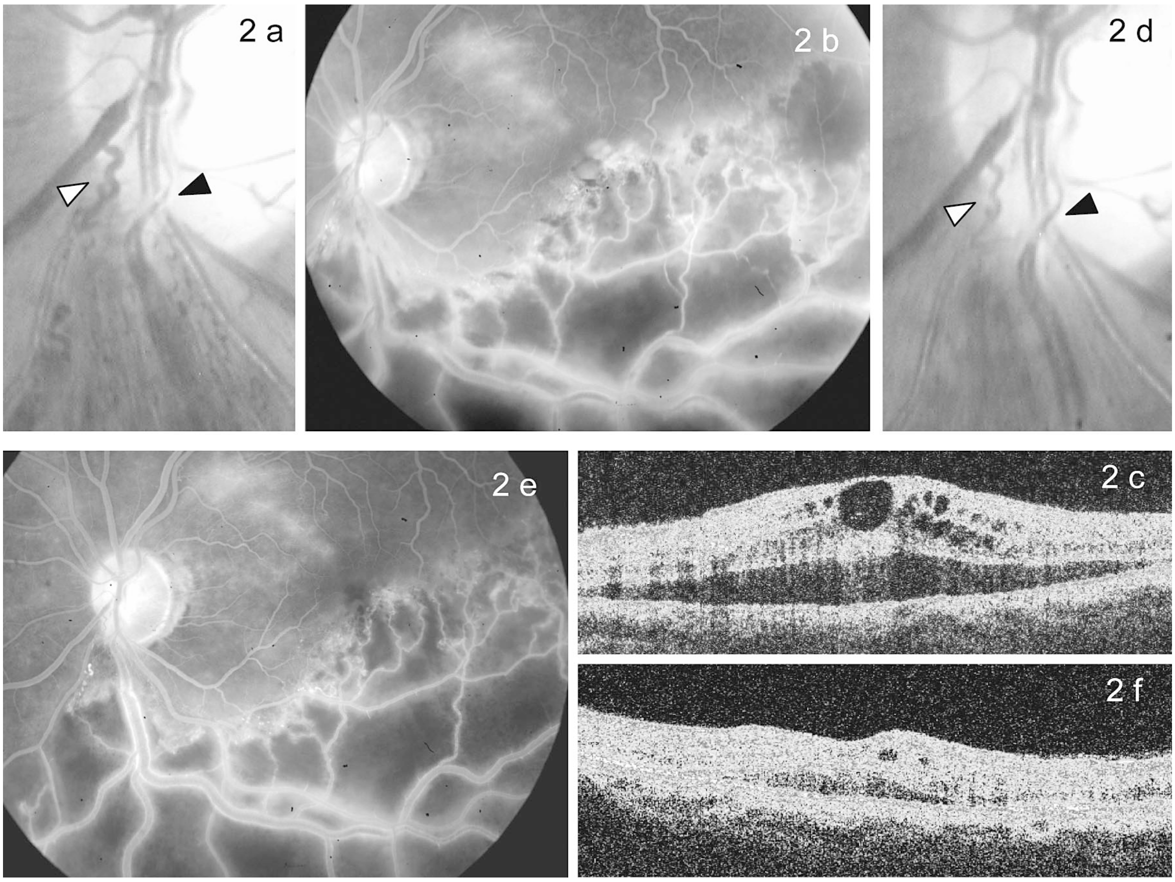

Fig. 2 Demonstrates fundus photographs, fluorescein angiography and OCT of the left eye of case 2
2a) Preoperative fundus photograph shows venule overlying arteriole at the crossing site with collateral vessel. Black arrowhead point to the occlusion site, white arrowhead point to the collateral vessel
2b) Preoperative fluorescein angiography shows occlusion of inferotemporal retinal vein with macular edema and non perfusion
2c) Preoperative OCT shows macular edema with increased foveal thickness
2d) Postoperative 1 week shows dilatation of the retinal vein at occlusion site (black arrowhead) and decreased collateral drainage (white arrowhead)
2e) Postoperative fluorescein angiography at 1 week shows retinal vein dilatation, decreased macular edema and improved retinal perfusion
2f) OCT taken 1 week after surgery shows decreased foveal retinal thickness

onset of BRVO[4]. The exact duration of the occlusion in BRVO patients is often unknown, especially when the fellow eye has normal vision. This may have an effect when considering whether treatment should be deferred or whether the occlusion is unlikely to have the potential for spontaneous improvement and treatment should be considered. Both of the presented cases had progressive visual decrease to 6/60 from severe macular edema, hemorrhage and ischemia with preoperative FA showed leakage, edema and non perfusion area in the macula. These were poor prognostic signs for spontaneous visual improvement and laser treatment may not be effective for restoring blood flow. The authors decided to perform AAS in both eyes in order to improve perfusion of the macula.

From the literature review, the artery was located anterior to the vein at the obstruction site in all except one previous sheathotomy studies. Yamaji et al[22] reported evaluation of sheathotomy for BRVO by fluorescein videoangiography and image analysis in 18 consecutive eyes, 5 eyes showing the venule overlying the arteriole, improvement in delay in the perfusion was observed in only one of the five. They discussed that the venule in all five eyes might be a collateral vessel, not a preexisting vessel, because the venule overlying the arteriole was small-sized and separation was technically difficult and incomplete for the small-sized venule. Contrary to the presented patients, both involved branch veins overlying arteries were not collateral vessels. Even though the authors could not separate A/V crossing in the first case and the authors did not attempt to separate A/V crossing in the second case, intraoperative dilatation, pulsation and restoration of downstream blood flow of the involved vein were observed with postoperative rapid improvement of visual acuity, clinical examination and fluorescein angiogram in both eyes.

The benefit of treatment for BRVO has to be compared with the natural course of BRVO that can have spontaneous improvement[5-7].The mechanism for spontaneous resolution of macular edema and ischemia may be the formation of collateral vessels[4]. However, the mechanism for improvement after surgical treatment for BRVO is uncertain. Some studies proposed that vitrectomy with posterior hyaloid removal[19,24,26,32,33] and internal limiting membrane removal[15,17,21] whether or not sheathotomy was performed could improve macular edema although macular traction is not usually detected in BRVO cases. Steffanson[33] suggested that vitrectomy improved the oxygen supply to the ischemic inner retina by way of fluid currents in the vitreous cavity. The other explanation may be the vascular endothelial growth factor that promote vascular leakage is more diffuse from the retina after vitrectomy.

In contrast, a recent study, by Lakhanpal et al[28] reported 25-gauge transvitreal limited arterio-venous-crossing manipulation without vitrectomy (LAM) with intraoperative manipulation of adventitia and conjoined vascular wall by lifting the proximal portion of the artery initially, followed by the portion distal to the crossing. Then, the artery was lifted at the crossing site, stretching but not severing the surgical connection between the vessels. This resulted in immediate restoration of venous blood flow in all 13 cases and visual improvement in 12 cases. Other than LAM, there was no other intraocular manipulation. None of the patients developed any signs of posterior vitreous detachment postoperatively.

A study by Han et al[27], however, proposed that incomplete separation of the retinal vessels might achieve results comparable with those reported by previous sheathotomy studies. The author's explanation is speculation that a configuration of the occluded vein as it curves beneath an overlying artery may result in turbulent flow and alteration of this configuration of the vessels by surgical displacement of the artery into a position anterior to the retina in some way may reverse a pathologic state. This may not explain both of the presented cases because veins still curve over the arteries pre- and postoperatively. The authors postulate that lysis of the sheath and manipulation of vein at and around the obstruction site may affect vein caliber and the thrombus at the occlusion site. Consideration of the hemodynamic consequences of flow by fluid dynamic principle of Poiseuille's law [Flow rate (Q) = $\pi r^4 P/ 8\eta L$ (r = tube radius, P = pressure difference between ends, η = fluid viscosity, L = pipe length)] that flow rate of fluid is proportional to the fourth power of the pipe radius, it means that 16 tubes can pass as much fluid as one tube twice its diameter. So if the diameter of the obstructed vessel is increased even by a little amount, the flow across the crossing site will be exponentially increased. The increase flow downstream and mechanical manipulation may dis-lodge the thrombus lead to continued reduction of upstream venous pressure, edematous retinal changes and hemorrhage and reduce downstream turbulence, endothelial damage and secondary thrombosis in the vein. This may imply the presented result of rapid improvement in both cases. The gradual improvement of macular edema and ischemia may be because of microcirculation improvement by collateral formation.

There are potential complications associated with this surgery including retinal tear or detachment, vitreous hemorrhage, retinal gliosis, nerve fiber layer defect, and accelerated nuclear sclerotic cataract[9-15, 18,20]. In the first case, retinal pigment epithelial alteration from phototoxicity was found postoperatively by FA but the patient did not complain of scotoma and in the second case, a small hemorrhage from a small collateral vessel was observed and stopped by increased intraocular pressure.

In the second case, the authors used OCT to display high-resolution (axial resolution, 10 microns), cross-sectional retinal images that objectively document pre-versus postoperative retinal thickness and the efficacy of surgical outcomes. Puliafito et al[34] indicated that OCT could be used as an objective, non-invasive, noncontact, sensitive means of imaging macular diseases, particularly macular edema. Spaide et al[35] studied using OCT in BRVO cases to detect minute disturbances within and beneath the retina areas of serous retinal detachment because of its sensitivity. The use of OCT in the authors second patient allowed us to directly quantify the postoperative improvement in macular edema.

This report of treatment for uncommon BRVO cases may give some additional information to consi-der about unclear mechanism of BRVO and sheatho-tomy. The results from the present report suggested that venous sheathotomy without separation of vessels for BRVO with venule overlying arteriole at the occlusion site may be a beneficial in the reduction of obstruction, restoring blood flow, reduce macular edema and improve VA in selected cases. Further study is necessary to understand more about the mechanism and also to evaluate optimal timing, indication and efficacy of treatment for BRVO.

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การรักษาโรคแขนงหลอดเลือดดำในจอประสาทตาอุดตันที่มีหลอดเลือดดำทับบนหลอดเลือดแดงโดยการผ่าตัดเยื่อหุ้มผนังหลอดเลือดดำและไม่แยกหลอดเลือดดำจากหลอดเลือดแดง

ณัฐวุฒิ รอดอนันต์, โสมนัส ถุงสุวรรณ

รายงานผู้ป่วย 2 รายที่เป็นโรคแขนงหลอดเลือดดำของจอประสาทตาอุดตัน (branch retinal vein occlusion) ที่มีหลอดเลือดดำทับบนหลอดเลือดแดงในตำแหน่งที่มีการอุดตันซึ่งพบน้อยกว่า 1% ผู้ป่วยทั้ง 2 รายมีระดับสายตาลดลงอย่างรวดเร็วเป็น 6/60 และได้รับการผ่าตัดเยื่อหุ้มผนังหลอดเลือด (sheathotomy) โดยไม่แยกหลอดเลือดดำออกจากหลอดเลือดแดง ขณะผ่าตัดสังเกตเห็นการเต้นของหลอดเลือดดำโดยหลอดเลือดขยายขนาดขึ้นอย่างชัดเจน ผู้ป่วยรายแรกมีระดับสายตาดีขึ้นเป็น 6/36 หลังผ่าตัด 1 วัน และดีขึ้นเป็น 6/9 และ 6/6 หลังการ ผ่าตัดที่ 2 และ 6 สัปดาห์ และคงที่ตลอด 12 เดือน ผู้ป่วยรายที่ 2 มีระดับสายตาดีขึ้น เป็น 6/24 หลังผ่าตัด 1 วัน และดีขึ้นเป็น 6/18 หลังการผ่าตัด1 สัปดาห์ และค่อยดีขึ้นเกือบเป็นปกติในการติดตามผลครั้งสุดท้ายที่ 2 เดือน จากการตรวจการไหลเวียนของเลือดโดยการฉีดสี Fluorescein angiography ของทั้ง 2 ตาพบว่าก่อนผ่าตัดมีการลดลงของเลือดที่มาเลี้ยงจุดรับภาพ ภายหลังผ่าตัดมีเลือดมาเลี้ยงเพิ่มขึ้น หลอดเลือดดำที่ทับบนหลอดเลือดแดงขยายขนาดขึ้นและจุดรับภาพบวมลดลง และจากผลการตรวจ optical coherence tomography (OCT) ใน ผู้ป่วยรายที่ 2 พบว่าหลังผ่าตัดจุดรับภาพบวมลดลงชัดเจน จาก 874 ไมครอน ก่อนผ่าตัด เป็น 420 ไมครอน หลังผ่าตัด 1 สัปดาห์

การรักษาโรคแขนงหลอดเลือดดำในจอประสาทตาอุดตันโดยการผ่าตัดเยื่อหุ้มผนังหลอดเลือดในตำแหน่งที่หลอดเลือดดำทับบนหลอดเลือดแดงโดยไม่แยกหลอดเลือดออกจากกันอาจทำให้ระดับสายตาดีขึ้นรวมทั้งจุดรับภาพยุบบวมลง