Polskie Towarzystwo Okulistyczne
www.pto.com.pl
KLINIKA OCZNA 2021, 123, 4: 210–214
Received: 13.11.2020 Accepted: 20.12.2020

CASE REPORT

Access this article online
Website: www.klinikaoczna.pl
DOI: https://doi.org/10.5114/ko.2021.111822

Swept-source optical coherence tomography angiography in the course of anti-VEGF treatment of choroidal neovascularization subsequent to birdshot chorioretinopathy

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ABSTRACT

The aim of the study was to present swept-source optical coherence tomography (OCT) angiography in a case of birdshot retinopathy complicated by choroidal neovascularization (CNV).

Material and methods: 59-year-old, HLA-A29 positive woman, treated for three years for birdshot chorioretinopathy with infliximab for systemic immunosuppression, presented to our clinic with decline in visual acuity in her left eye. Swept-source OCT and swept-source OCT angiography were performed additionally to fluorescein angiography and indocyanine green angiography. 0.05 ml of bevacizumab treatment was administered monthly for five months. Anti-VEGF treatment was administered with improvement in visual acuity, but without complete regression of CNV.

Results: At presentation visual acuity was 0.4 in her right eye and 0.1 in left eye. One month after anti- VEGF administration, we observed improvement of visual acuity to 0.3, with slight regression of the branched network on swept-source OCT angiography. After the second anti-VEGF injection visual acuity in this eye improved to 0.4. The fovea contour normalized and the hyperreflectivity in the swept-source OCT area above RPE corresponding to branched CNV disappeared.

Conclusions: Swept-source OCT angiography revealed two findings. First, we observed numerous hyporeflective spots visible in the superficial and deep retinal layer, but most pronounced in the choriocapillaries. Those are probably corresponding with the activity of the disease. Second, we observed CNV in one eye.

KEY WORDS: anti-VEGF treatment, birdshot chorioretinopathy, HLA-A29, swept-source OCT angiography, SS-OCTA.

INTRODUCTION

Birdshot chorioretinopathy is a rare bilateral autoimmune disorder, strongly associated with human leukocyte antigen (HLA) A29. The clinical picture consists of multiple cream-color spots spreading radially from the optic nerve into the periphery with coexisting vitreous haze. This disease can be complicated by several fovea abnormalities, most often macular edema and retinal vasculitis [1]. A less common complication of this disease is choroidal neovascularization (CNV), previously observed in about 6-14% of cases [2]. The most reasonable approach to treat CNV in the course of birdshot chorioretinopathy is anti-VEGF therapy. However, diagnosing CNV and monitoring therapy can be complicated with standard imaging: optical coherence tomography (OCT), fluorescein angiography (FA), indocyanine green angiography (ICG).

Swept-source OCT angiography (SS-OCTA) is a novel noninvasive technique presenting movement of cells in retinal and choroidal tissue at particular levels of vasculature. Most systems use software that indicates the level of superficial and deep retina vessel plexus, avascular zone and choriocapillaries. CNV is observed in SS-OCTA as a branched network of vessels.

CASE PRESENTATION

A 59-year-old, HLA-A29 positive woman, treated for three years for birdshot chorioretinopathy with infliximab, presented with decline in visual acuity in her left eye (0.1 Snellen). Visual acuity in the fellow eye was 0.4 Snellen (Figure 1). Additionally, the woman underwent laser treatment of retinoschisis in the lower periphery in both eyes. SS-OCT A (Triton, Topcon, Japan) revealed a branched network corresponding to CNV (Figure 2B, white arrow). In addition,

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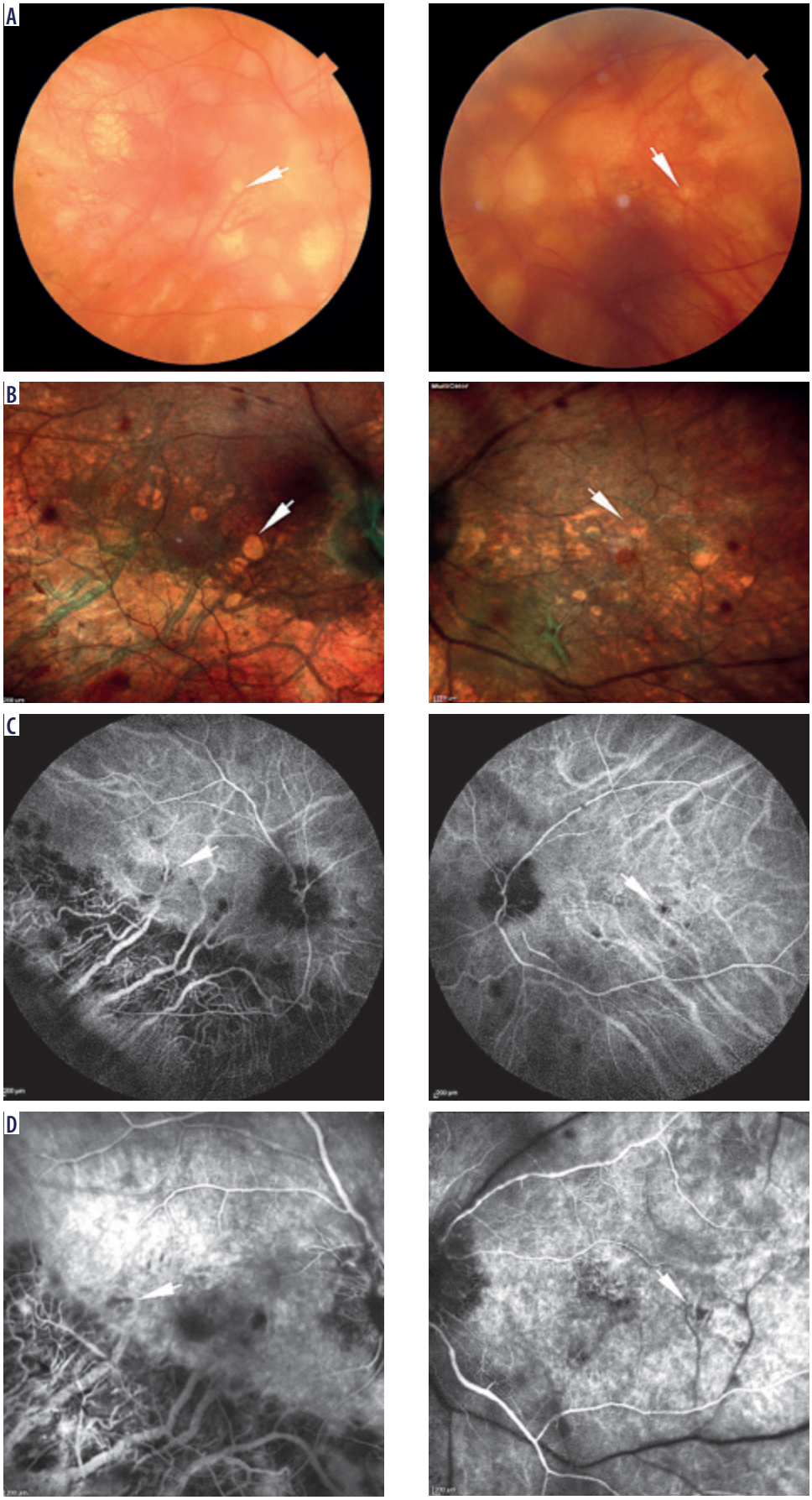

Figure 1. A) Fundus image of a 59-year-old woman with birdshot chorioretinopathy. Cream-colored lesions are visible as well as stage one vitreous haze. **B**) Multicolor image (Spectralis, Heidelberg Engineering) of the same patient. **C**) Indocyanine green angiography of the same patient. **D**) Fluorescein angiography of the same patient

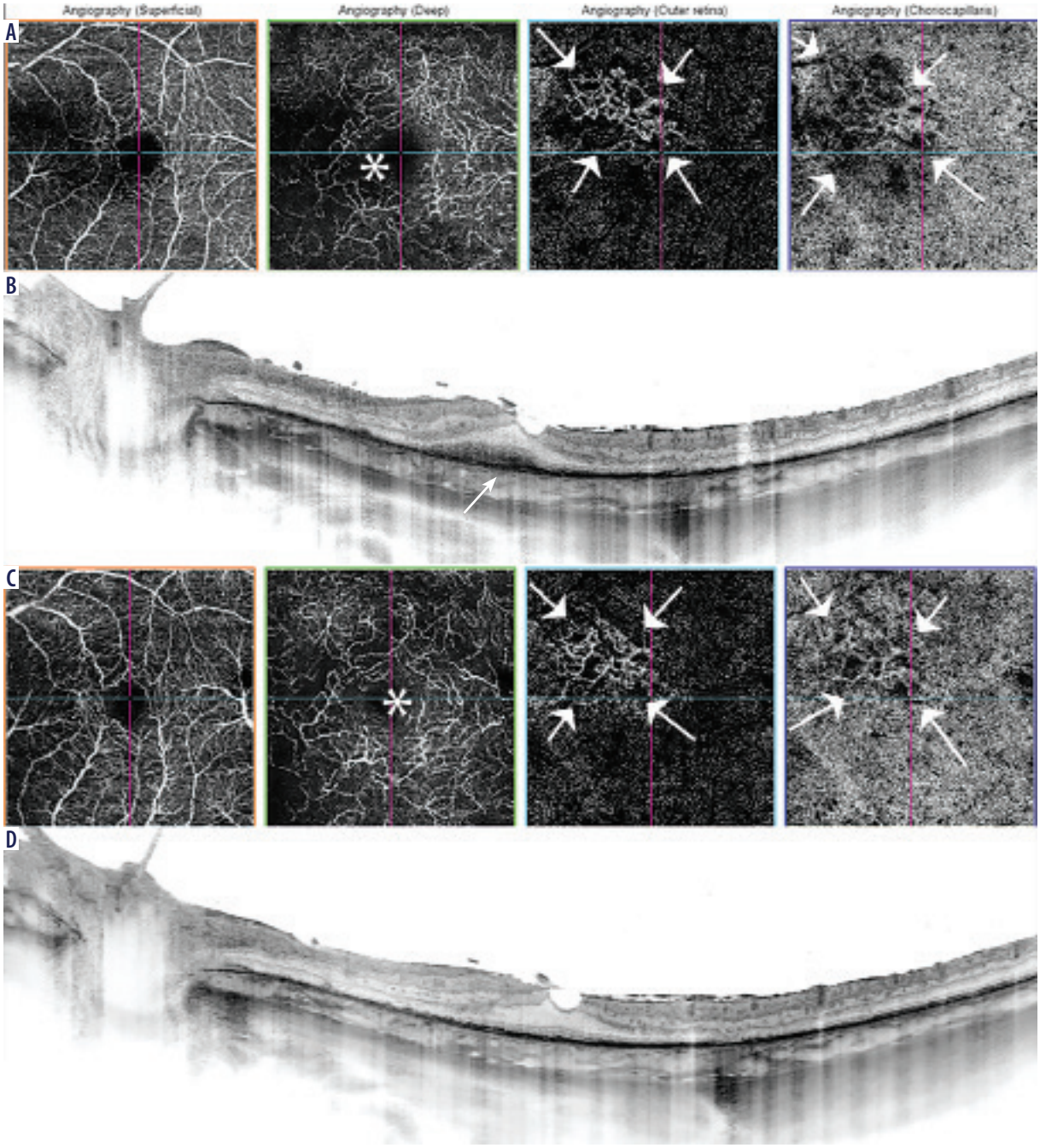

Figure 2. A) Swept-source OCT angiography (SS-OCT A) at presentation of choroidal neovascularization (CNV) in the course of birdshot chorioretinopathy. From left: Superficial retinal vessel layer, deep retinal vessels layer, avascular zone, choriocapillaries. CNV is visible in the avascular zone and choriocapillaries (white arrows). **B**) Swept-source OCT (SS-OCT) – type two neovascular membrane is visible as hyperreflective elevation above the retinal pigment epithelium (black arrow). No subretinal or intraretinal fluid was noted. **C**) SS-OCT A after two anti- EGF injections. Branched neovascularization is still visible at the level of the avascular zone and choriocapillaries. From left: Superficial retinal vessel layer, deep retinal vessels layer, avascular zone, choriocapillaries. CNV is visible in the avascular zone and choriocapillaries (white arrows). **D**) SS-OCT after two anti-VEGF injections shows complete restoration of the fovea contour. CNV is no longer visible on SS-OCT. The star indicates the deep retinal vessel layer

we observed decreased density of vessels in deep retina layers (Figure 2A, middle left, star). Images of the periphery of both eyes indicated several hyporeflective spots (Figure 3). Some of those spots corresponded to hypofluorescent spots on ICG, but many of them were visible only in SS-OCT A (Figure 3, white arrows). Those spots were visible at the level of superficial retinal vessels, and even more numerous at the level of choriocapillaries (Figures 3, 4). 0.05 ml of bevacizumab treatment was administered monthly for five months.

We noted an improvement of visual acuity to 0.4, with slight regression of the branched network on SS-OCT A (Figure 2C, D).

A new finding established by us using SS-OCT A displayed hyporeflective areas not only at the level of chorio-

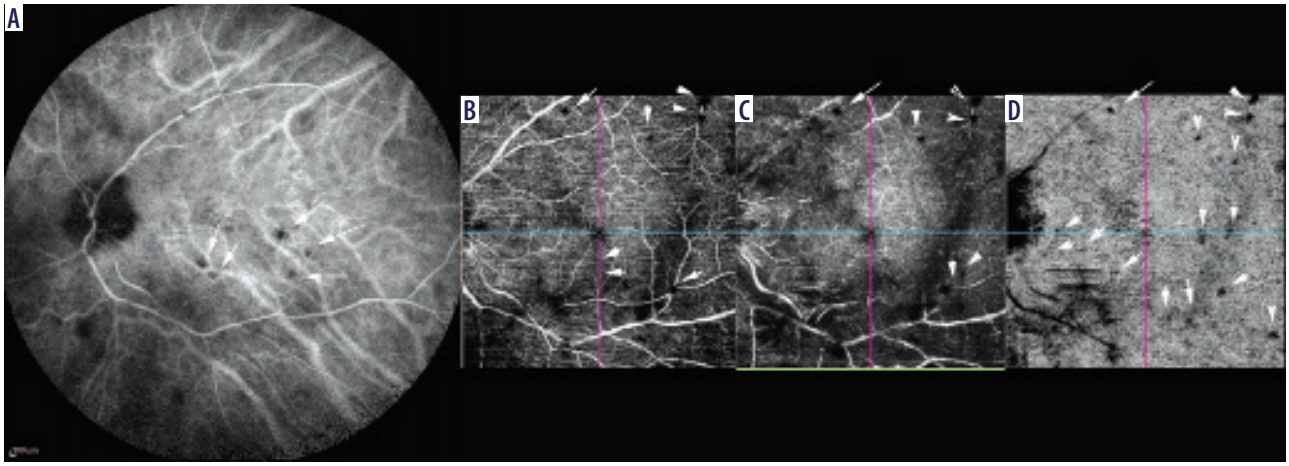

Figure 3. Indocyanine green angiography and corresponding swept-source OCT angiography (SS-OCT A). Hypocyanine spots and hyporeflective spots are indicated with white arrows. **A**) Indocyanine green angiography of the left eye. Hypocyanine spots corresponding to disease activity are marked with white arrows. **B**) SS-OCT A at the level of superficial retinal vessel plexus. Hyporeflective spots are marked with white arrows. It is visible that only a few correspond to indocyanine angiography. **C**) SS-OCT A at the level of deep retinal vessel plexus. Hyporeflective spots are marked with white arrows. It is visible that only a few correspond to indocyanine angiography, but many correspond to SS-OCT A at the level of the superficial retinal vessel plexus. **D**) SS-OCT A at the level of choriocapillaries. Hyporeflective spots are marked with white arrows. It is visible that they are more numerous than in other layers and when compared to indocyanine angiography

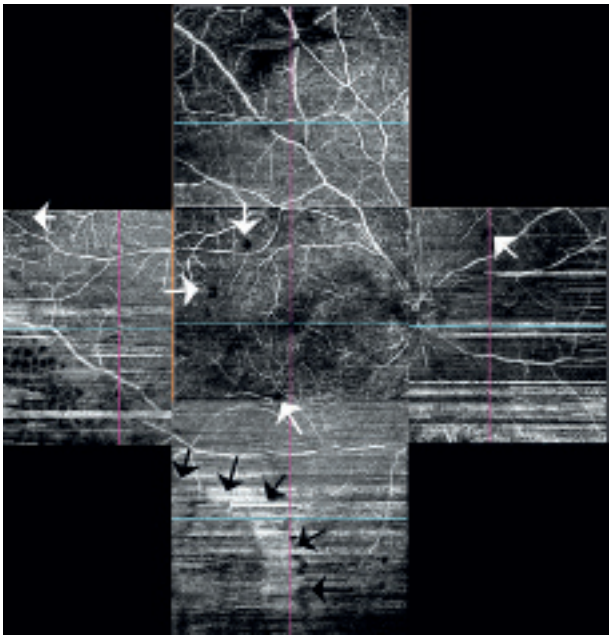

Figure 4. Montage swept-source OCT angiographic image of superficial retinal vessel plexus in the right eye of a patient with birdshot chorioretinopathy. Hyporeflective spots, probably indicating the activity of the disease, are visible (white arrows). Black arrows indicate laser coagulation at the margin of retinoschisis

capillaris, but also at superficial and deep retina layers. Some of those areas correspond precisely to the localization of hypoperfusion sites of the choroid. However, hyporeflective areas are more numerous at the level of the choroid. It is rather improbable to label this as an artifact, as they were visible in both eyes in the entire periphery (imaged with different angles).

Reduced vessel density, as observed in birdshot [3], has long been thought to correlate with CNV formation in uveitis patients [4]. To the best of our knowledge, there are no data on SS-OCT A in the course of CNV treatment of birdshot choroidopathy. In our study, only minor changes in the area of CNV were visible in SS-OCT A after a series of anti-VEGF injections in spite of improvement in visual acuity and improvement in SS-OCT.

CONCLUSIONS

SS-OCTA is a useful tool for monitoring the treatment of CNV complicating birdshot chorioretinopathy and is able to visualize peripheral lesions in this disease. Further studies are needed to ascertain whether it could compliment or even replace ICG in monitoring immunosuppression therapy.

Earlier OCTA based studies described areas of reduced choroidal flow corresponding to the cream-colored dots visible on the fundus [5, 6]. Large choroidal vessels traversing or bordering those areas of choriocapillary hypoperfusion were also observed in earlier ICG and EDI-OCT based studies [7].

Retinal vascular changes in birdshot chorioretinopathy are not well studied. Dye leakage on FA might correspond to thickening of the retina on OCT [8]. We observed reduced vessel density and vessel loops in the deep retinal vessels layer, which were also noted in SD-OCTA based studies.

As a new finding, we established that SS-OCTA showed hyporeflective areas not only at the level of choriocapillaris, as described before [9], but also in the superficial and deep retina layers. Those areas correspond precisely to the localization of hypoperfusion sites of the choroid. We dismissed the idea that they could be artifacts as they were visible in both eyes in the whole periphery (imaged at different angles).

Reduced vessel density has long been thought to correlate with CNV formation in uveitis patients [4] and could be the cause of CNV in our patient.

Information on the treatment of CNV complicating birdshot chorioretinopathy in the literature is limited to case reports. Shanta and coworkers presented four such cases, achieving a statistically significant improvement in visual acuity. They reported a mean of 0.1 events of CNV per patient per year [10]. Similarly, Brue and coworkers presented a young woman with bilateral CNV treated with a single injection of bevacizumab and triamcinolone and photodynamic therapy in one eye, while the other eye responded well to triamcinolone injections [11]. To the best of our knowledge there are no data on SS-OCTA in the course of treatment of birdshot choroidopathy. In our study, despite improvement in visual acuity, and improvement in SS-OCT, no changes in the area of CNV were visible in SS-OCTA after two anti-VEGF injections.

In conclusion, CNV in birdshot chorioretinopathy responds well to anti-VEGF treatment, but continuous injections might be mandatory. SS-OCT might be advised for monitoring of anti-VEGF treatment as it demonstrated a branched network of the CNV. Additionally, this technique might be considered to monitor immunosuppressive treatment as it demonstrates peripheral lesions in birdshot chorioretinopathy at different levels in SS-OCT A. At the level of choriocapillaris they are even more numerous than in ICG.

DISCLOSURE

The authors declare no conflict of interest.

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