Serpiginous choroidopathy present as choroidal neovascularisation

Serpiginous choroidopathy is an insidious, relentlessly progressive, idiopathic inflammatory disease affecting the retinal pigment epithelium and inner choroid. Choroidal neovascularisation (CNV) is a well recognised late complication of serpiginous choroidopathy in 10–25% of affected patients. In all previously reported cases CNV was recognised at the time of or after the diagnosis of serpiginous choroidopathy was established. We report a patient presenting with CNV who subsequently developed clinical findings characteristic of serpiginous choroidopathy.

Case report
A 31 year old man presented with decreased vision in his right eye in July 1997. Examination revealed acuities of 20/40 right eye and 20/20 left eye with normal anterior segments. The right fundus showed subretinal fluid and haemorrhage adjacent to the disc (Fig 1A). The left eye showed an irregularity superior to the optic disc (Fig 1B). The vitreous and fundi were otherwise normal bilaterally. Fluorescein angiography (Fig 2A, B) revealed peripapillary choroidal neovascular membranes in both eyes that were treated with argon laser photocoagulation. In April 1998 and February 1999 the left eye required photocoagulation for recurrent peripapillary CNV. Evaluation for floaters in February 2000 revealed 1+ vitreous cells and new lesions in the left eye.

Examination at the National Eye Institute in April 2000 revealed acuities of 20/40 right eye and 20/16 left eye with normal anterior segments. The vitreous contained trace cells without haze bilaterally. The right fundus showed a large peripapillary chorioretinal scar. The left fundus revealed a chorioretinal scar superior to the disc and two yellow, irregularly circumscribed, deep macular lesions (Fig 3A, B). The retinal vessels and discs were normal and no subretinal fluid, haemorrhage, or macular oedema was noted in either eye.

Fluorescein angiography revealed early hypofluorescence and late hyperfluorescence corresponding to the macular lesions in the left eye (Fig 3C, D) with no evidence of CNV in either eye. Laboratory studies were non-diagnostic. A diagnosis of serpiginous choroidopathy was made based on the clinical and fluorescein characteristics of the macular lesions in the left eye.

Comment
CNV in serpiginous choroidopathy is associated with a poor visual prognosis. In a small study CNV was reported to develop within 16 months of the serpiginous diagnosis. In a larger retrospective study of 53 serpiginous patients active CNV was found in three patients at the time of initial diagnosis and in three others within 2–17 months. Our patient differs from those previously reported in that he was diagnosed and treated for idiopathic CNV before the recognition of clinical findings.
Optic neuritis in anti-GQ1b positive recurrent Miller Fisher syndrome

Only five cases of optic nerve involvement in Miller Fisher syndrome (MFS) have been documented in the literature. This report further confirms that optic neuritis may be seen in anti-GQ1b positive MFS.

Case report

This 23 year old woman presented with acute blury vision, diplopia, and pain with eye movement. Her visual acuity was 20/20 in both eyes, but her left disc remained oedematous. She had bilateral sixth nerve palsies, nystagmus in all gazes, and left optic disc oedema. After 1 week her visual acuity improved to 20/20 in both eyes, but her left disc remained oedematous. She developed exacerbation of gait ataxia, and she noticed a decrease in her ability to touch in one arm. She also had tingling in her hands and feet and decreased lower extremity vibratory sensation. Her mental status was normal throughout her illness. She was not taking any drugs. A magnetic resonance image (MRI) of the brain and entire spine and MR venogram were all normal. Her cerebrospinal fluid (CSF) opening pressure was 190 mm H2O. Her CSF protein was elevated at 70 mg/dl, but CSF glucose and cell count were normal. CSF VDRL, Gram stain, routine bacterial, viral, and fungal cultures were all negative. Anti-GD1a, anti-GM1, and anti-GQ1b antibodies were all negative. After 1 week of treatment with intravenous methylprednisolone, her visual acuity improved to 20/20 in both eyes, and optic disc oedema markedly decreased. She continued to have blurred vision, diplopia, and pain with eye movement, dyschromatopsia, and optic disc oedema that resulted in good visual recovery are all indicative of the diagnosis of optic neuritis. Since high titres of anti-GQ1b antibody can cross the blood-brain barrier, the optic disc oedema in this patient could represent central anti-GQ1b IgG optic neuritis, or a CNS immune inflammatory demyelination. Furthermore, her ipsilateral delayed P100 latency is consistent with a pre-chiasmal demyelinating optic neuritis. In addition to her optic neuritis, this patient concomitantly demonstrated the classic features of MFS which are the acute onset of external ophtalmoplegia, ataxia of the cerebellar type, and the loss of peripheral sensory and reflexes. MFS is considered a variant of Guillain-Barré syndrome (GBS) because some patients who present with MFS progress to GBS. High titres of anti-GQ1b IgG antibodies are present in 80% to 100% of patients with MFS. MFS may be immunologically differentiated from GBS by the presence of anti-GQ1b and anti-GM1 antibodies. Although both anti-GD1a IgG and anti-GM1 IgG are associated with GBS, anti-GM1 IgG is present in patients with typical MFS who have limb weakness, as in this patient. As further evidence linking this antibody to MFS, the decrease in anti-GQ1b antibody levels after plasmapheresis correlated with the clinical recovery in this patient. Therefore, the elevated titres of anti-GQ1b and anti-GM1 antibodies, along with the clinical trial of optalmoplegia, areflexia, and ataxia in this patient all support the diagnosis of MFS, and not GBS.

In rare cases, MFS has been known to recur. This patient presented with a relapse of similar clinical features 6 months after her initial episode. In the study done by Chida et al., patients with recurrent MFS appeared to have similar HLA typing characteristics as the non-recurring ones. Both types of MFS may share HLA-DR2 and Cw3 alleles, but the frequency of HLA-DR2 was slightly higher in the patients with recurrent MFS. Therefore, this patient’s HLA-DR2-positive status may have been a risk factor for her recurrence of MFS.

This case report emphasises that optic neuritis may be a central nervous system feature that should be recognised as part of the Miller Fisher syndrome. The presence of both anti-GQ1b IgG and anti-GM1 IgG in this patient provides immunological evidence supportive...
of typical MFS. The delayed P100 latency in her VEP also provides electrophysiological evidence that the optic nerve is affected in anti-GQ1b antibody-positive MFS. Furthermore, this is the first documented case known to the author of optic neuritis in the recurrent subtype of MFS which is associated with a higher frequency of the HLA-DR2 allele.

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Ocular myasthenia gravis and inflammatory bowel disease: a case report and literature review

Myasthenia gravis has been reported to be associated with both ulcerative colitis (UC) and Crohn’s disease (CD). The link between inflammatory bowel disease (IBD) and myasthenia gravis (MG) is thought to be related to the production of autoantibodies. Myasthenia gravis is also associated with other autoimmune diseases including alopecia, lichen planus, vitiligo, and systemic lupus erythematosus.

Similarly, IBD frequently presents with other autoimmune disorders. One study demonstrated a 9.4% prevalence of autoimmune disorders in patients with UC including sclerosing cholangitis, thyroid disorders, vitiligo, insulin dependent diabetes mellitus, thyroid disease, pernicious anaemia, scleroderma, and seropositive rheumatoid arthritis. Despite the association between MG and other autoimmune disorders, there are relatively few reports of ocular findings as the presenting sign of MG in patients with IBD.

Case report

A 21 year old African-American male, with a medical history of biopsy proved ulcerative colitis diagnosed in 1995, focal segmental glomerular sclerosis determined by renal biopsy in 1995, and primary sclerosing cholangitis determined by liver biopsy in 2000 presented to the neuro-ophthalmology service with complaints of binocular diplopia and ptosis of the left upper eyelid. Both the diplopia and the ptosis were better in the morning and worsened during the course of the day. His ulcerative colitis had been in remission for the past 5 years without medication.

Best corrected visual acuity was 20/25 in each eye. The external examination revealed ptosis of the left upper eyelid that worsened in sustained upgaze. He had limited extraocular motility in all fields of gaze (Fig 1). The remainder of the neuro-ophthalmic examination was normal and he had no difficulty with speech or swallowing.

Laboratory evaluation revealed a positive acetylcholine receptor antibody and normal thyroid function studies. There was no evidence of a thymic mass on magnetic resonance imaging of the chest.

The patient returned to the emergency room 1 week later with difficulty swallowing and shortness of breath. He was hospitalised for plasmapheresis and upon discharge treated with immunotherapy.

Comment

Autoimmune disorders, including MG, occur more frequently in UC than in CD. It is not clear how many other cases of IBD manifested with ocular presentations as the initial finding of MG as in our case report. Our literature review revealed only one other purely ocular presentation of myasthenia associated with ulcerative colitis; however, details of the ocular examination were not included.

Another report, of a 21 year old woman with a 3 year history of Crohn’s disease, documented diplopia and unilateral ptosis as the initial findings of MG. She was found to have acetylcholine receptor antibodies and her ocular findings improved with pyridostigmine.

Because of the relatively few reports of ocular myasthenia in patients with IBD we reviewed the English literature and found four additional reports of MG in patients with IBD. Based on these four reports and the three (including the present report) with ocular MG in patients with IBD (Table 1), the mean duration of IBD before the diagnosis of MG was 10 years.

Autoimmune dysregulation is the central defect in both MG and IBD. Both IBD and MG may be associated with an elevated carcinoembryonic antigen (CEA) and decreased peripheral lymphocyte counts that subsequently normalise following thymectomy. Some studies have shown abnormal thymic involution and the presence of an abnormal ratio of T suppressor to T helper cells in both MG and UC, while others have noted a decline in suppressor T cells and an increase in

Table 1

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age (years)</th>
<th>Sex</th>
<th>IBD</th>
<th>Duration of IBD before diagnosis of MG (years)</th>
<th>AchR antibody reactivity</th>
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<tr>
<td>Miller et al., 1971</td>
<td>35</td>
<td>Male</td>
<td>UC</td>
<td>13</td>
<td>Unknown</td>
</tr>
<tr>
<td>Tan et al., 1974</td>
<td>38</td>
<td>Male</td>
<td>UC</td>
<td>12</td>
<td>Unknown</td>
</tr>
<tr>
<td>Martin et al., 1991</td>
<td>63</td>
<td>Male</td>
<td>CD</td>
<td>15</td>
<td>Positive</td>
</tr>
<tr>
<td>Gowen-Rousseau et al., 1993</td>
<td>27</td>
<td>Female</td>
<td>UC</td>
<td>10</td>
<td>Positive</td>
</tr>
<tr>
<td>Finnie et al., 1994</td>
<td>21</td>
<td>Female</td>
<td>CD</td>
<td>3</td>
<td>Positive</td>
</tr>
<tr>
<td>Lassos et al., 1995</td>
<td>11</td>
<td>Male</td>
<td>CD</td>
<td>9</td>
<td>Unknown</td>
</tr>
<tr>
<td>Present report</td>
<td>21</td>
<td>Male</td>
<td>UC</td>
<td>7</td>
<td>Positive</td>
</tr>
</tbody>
</table>

IBD = inflammatory bowel disease, MG = myasthenia gravis, AchR = acetylcholine receptor, UC = ulcerative colitis, CD = Crohn’s disease.

Figure 1

External photograph shows ptosis of the left upper eyelid, restriction of all extraocular movements of the left eye, and an elevation and adduction deficit of the right eye.
immature helper T cells suggesting migration without normal maturation.\(^3\)\(^4\)

The immunological link between MG and IBD is highlighted by two reports of patients undergoing surgical treatment. One report of a patient with both MG and CD documented improvement in perineal and perianal disease following thymectomy for severe uncontrolled MG.\(^5\) Another patient with both MG and UC demonstrated regression of the myasthenia following proctectomy.\(^6\)

Although the simultaneous occurrence of these two autoimmune disorders is uncommon, it is important to understand that ocular findings may be the initial manifestation of MG in patients with IBD.

The authors have no proprietary interest in any contents of this manuscript.

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References

Magnetic resonance imaging findings in malignant melanoma of the lacrimal sac

A case of primary malignant melanoma of the lacrimal sac is presented. This is the first report of the preoperative magnetic resonance imaging (MRI) findings of malignant melanoma of the lacrimal sac.

Case report

A 54 year old Chinese woman was referred to an ophthalmologist complaining of a 6 month history of left sided bloody tears and epistaxis. She had a firm, non-tender left medial canthal swelling, and syringing revealed left nasolacrimal duct (NLD) obstruction. Ocular and periorbital examination was otherwise normal. A dacryocystogram (DCG) demonstrated a filling defect in the lacrimal sac with NLD obstruction.

An ENT opinion was sought, and nasal examination revealed left sided septal deviation, with no obvious cause for the epistaxis. Computed tomography (CT) of the head and orbits demonstrated a left lacrimal sac lesion extending into the NLD with proximal dilation of the duct and no apparent bone erosion. An incisional biopsy of the lacrimal sac (Fig 1B) under frozen section control, and paraffin sections, confirmed malignant melanoma.

An full medical review, including MRI of the chest and abdomen, and liver function tests, excluded tumour elsewhere. However, abdominal MRI and ultrasound revealed a co-incidental polycyclic liver.

Three weeks after biopsy, a wide local excision including the medial upper and lower eyelids, dacryocystectomy and medial maxillectomy was performed. A tumour, confined to the sac, and invasion through the medial wall of the upper NLD, into the lateral wall of the nose, and apposing nasal septal mucosa, was seen peroperatively and confirmed histologically.

She underwent postoperative adjuvant radiotherapy (55 grays) and to date, 4 months later, remains well.

Comment

Malignant melanoma of the lacrimal sac is rare accounting for 3% of lacrimal sac tumours.\(^7\)\(^8\) It has an unfavourable prognosis compared with other causes of lacrimal sac tumour, and is considered more aggressive than cutaneous malignant melanoma.\(^7\)\(^8\) Response to treatment is generally poor, with up to 80% of cases recurring within 2 years.

Radiological features of lacrimal sac tumours include filling defects on DCG and mass lesions on CT.\(^7\) However, to the authors’ knowledge, this is the first report of the MRI findings of malignant melanoma of the lacrimal sac. Owing to the paramagnetic properties of melanin, malignant melanoma appears hyperintense on T1 weighted imaging, and hypointense on T2 weighted imaging.\(^4\) A study of six mucosal melanomas of the head and neck found that on T1, five lesions were hyperintense and one was isointense.\(^9\) On T2, five were of mixed intensity and one was iso-intense. They concluded that hyperintensity on T1 of mucosal melanomas was characteristic but not universal.

The majority of malignant lacrimal sac tumours are epithelial in origin.\(^4\) Imaging features suggesting malignancy include invasion of bone, rapid growth, and irregular margins with skin fixation. On MRI, the majority of epithelial tumours have intermediate signal intensity on T1 and high T2 signal intensity. High tumour cellularity is associated with intermediate to low T2 signal intensity.\(^4\)

High signal intensity on T1 is not specific for malignant melanoma. Subacute haemorrhage caused by the presence of methaemoglobin is more likely and although melanoma may undergo intratumoral haemorrhage, other tumours with a tendency to bleed may also be seen. Small cell lung carcinoma, choriocarcinoma, and renal cell carcinoma metastases. Less likely causes include fat containing tumours (lipoma, dermoid, and teratoma).

Figure 1 (A) Coronal CT scan demonstrating a solid mass of the left lacrimal sac with proximal dilation of the nasolacrimal duct (arrow). (B) Incisional biopsy with lacrimal sac opened and melanoma visible.

Figure 2 (A) T1 weighted sagittal MRI demonstrating intermediate signal intensity mass lesion of the lacrimal sac and proximal nasolacrimal duct (arrow). (B) T2 weighted axial MRI demonstrating intermediate signal intensity mass lesion of the left lacrimal sac (arrow).
requiring MRI fat suppression methods, \(^1\) paramagnetic material (manganese, iron, and copper), and very high (non-paramagnetic) intratumoral protein concentration.

MRI has been reported as a useful investigative tool in the assessment of macular disease owing to its ability to delineate soft tissues. Intravenous and intracanalicular gadolinium adds useful information on lesion enhancement and lacrimal apparatus structure and function. \(^2\) The predictive value of MRI for lacrimal sac melanoma, however, appears to be variable. Hyperintensity on T1 relies on the paramagnetic properties of melanin, the presence of which is variable in amelanotic melanoma. This is supported by our case, where only moderate T1 hyperintensity with contrast enhancement was demonstrated.

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References


Photodynamic therapy for recurrent myopic choroidal neovascularisation after limited macular translocation surgery

Limited macular translocation (LMT) is one of the treatment options for subfoveal choroidal neovascularisation (CNV) resulting from pathological myopia. \(^6\) The fundamental surgical principle involves the transposition of the foveal neurosensory retina to a new site with more healthy underlying retinal pigment epithelium. \(^4\) Direct laser photocoagulation is usually employed as an adjunct measure in eradicating the original CNV after the surgery. It has been observed that geometrically sizeable translocation is a prerequisite for a long term surgical success. \(^5\) The degree of translocation is, however, not often predictable and any ineffective displacement may render the subsequent laser photocoagulation extremely difficult or even impossible to perform. \(^3\) As a result, the recurrent or persistent CNV intruding the newly relocated fovea may jeopardise the final visual outcomes. \(^3\) Photodynamic therapy (PDT) may be considered a viable adjunct treatment option in such circumstance.

Case report

A 41-year-old woman with pathological myopia of \(-11.0\) dioptres in both eyes presented with a subfoveal CNV and subretinal haemorrhage in her right eye in July 2000. The best corrected visual acuity (BCVA) was 20/200 in her right eye and 20/30 in her left eye. LMT with superotemporal 6 mm scleral imbrication was performed in July 2000. The operation was uneventful and an inferior displacement of the fovea by 600 μm was achieved. The CNV however, was still located in the vicinity of the juxtafoveal area and therefore laser photocoagulation, bearing the potential risk of late creeping scar, was not suggested. At 4 months postoperative visit, her left BCVA was 20/200 and the original CNV became more fibrotic with minimal leakage upon fluorescein angiography. Nevertheless, she came back at 5 months with a return of metamorphopsia and a drop in her right vision from 20/200 to 10/200. Dilated fundus examination showed a tiny patch of submacular haemorrhage in direct continuity with the old fibrotic scar (Fig 1A). Fluorescein angiogram of the early phase demonstrated a fresh recurrent CNV budding out from the original CNV. The incidence of persistent or recurrent CNV after limited LMT has been reported to be 40% and 35% respectively in age related macular translocation and being 21% and 14% respectively in pathological myopia. \(^6\) However, the surgical techniques are demanding and the potential complications are not unusual. One of the late postoperative visually important complications is recurrence of the CNV and this is partially caused by an ineffective translocation of the fovea or a large lesion size of CNV. \(^5\) The vision remained stable at 20/200 in the latest visit at 24 months after the PDT.

Comment

It has been shown that significant visual improvement may be achieved by LMT for the treatment of subfoveal CNV associated with age related macular degeneration (AMD) or pathological myopia. \(^6\) However, the surgical techniques are demanding and the potential complications are not unusual. One of the late postoperative visually important complications is recurrence of the CNV and this is partially caused by an ineffective translocation of the fovea or a large lesion size of CNV. \(^5\) The incidence of persistent or recurrent CNV after limited LMT has been reported to be 40% and 35% respectively in age related macular translocation and being 21% and 14% respectively in pathological myopia. \(^6\) Not many treatment options are available once the fovea is involved. Viable surgical options including repeated LMT, full 360 degree retinotomy MT, or submacular surgery may be considered but the surgical risk may be inadvertently higher in the redetracement of the neurosensory retina. PDT induces a selective thrombosis of the abnormal CNV and has been proved to be an effective treatment in preventing a significant loss of vision in patients with CNV secondary to AMD or pathological myopia. \(^6\)
Its clinical indications and applications are expanding. Its minimal invasiveness and clinical efficacy make it a safer and visually desirable supplementary treatment in recurrent CNV after LMT. In our patient, the complete closure of CNV was achieved with concomitant vision improvement after a single session of PDT without evidence of recurrence at 24 months.

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References

Acquired Glanzmann’s thrombasthenia causing prolonged bleeding following phacoemulsification

Phacoemulsification under topical anaesthesia using clear corneal incision is not a challenging procedure for the haemostatic system. In patients with known bleeding dia-

Command

The patient described had uncontrollable bleeding for 36 hours following a procedure, which is generally considered to be safe in patients with a bleeding disorder. She developed bleeding from the conjunctival site where the surgeon grasped the conjunctiva during certain stages of the procedure. One would usually not expect any significant bleeding from this site; however, in a patient with compromised haemostasis the bleeding may be prolonged. Although the bleeding was no more than a gentle ooze at any point in time it was persistent enough for 36 hours before the topical haemostatic material Surgicel had been put to use. The consequences of an intraocular bleed may have seriously threatened her sight.

We are not aware of any reports of the use of Surgicel in ophthalmic surgery. All reports of its use are in other fields of surgery. This material is supposed to swell up with blood and form a gelatinous mass that aids in the formation of clot. It acts as a haemostatic adjunct. The exact mode of its action in this patient with antilatelet antibodies is unclear. Our experience shows that oxidised regenerated cellulose (Surgicel) may have a role in ophthalmic surgery especially in lenticular and orbital surgery, when faced with bleeding that is difficult to stop. Various cautionary tales associated with use of Surgicel have been reported. Our report suggests that in the presence of a severe bleeding disorder, clear corneal phacoemulsification under topical anaesthesia may not be totally safe. When performing such a procedure in a patient with known bleeding disorder it may be safest to take all the necessary precautions in consultation with a haematologist to avoid a serious bleed that may be sight and life threatening. There may be a role for haemostatic agents like Surgicel.
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Propionibacterium acnes endophthalmitis diagnosed by microdissection and PCR

Although Propionibacterium acnes, a Gram positive anaerobic bacillus, is the most commonly identified cause of delayed onset postoperative endophthalmitis, routine vitreous cultures are frequently inadequate for its diagnosis. This case describes the utility of the histopathological technique of microdissection and polymerase chain reaction (PCR) for the diagnosis of delayed postoperative endophthalmitis.

Case report

A 78 year old man with a history of vitreous floaters, a coronary bypass, and aortic valve replacement underwent an uncomplicated cataract extraction with intraocular lens (IOL) implantation in the right eye. Three months later, he developed increasing floaters in the right eye and was diagnosed with vitritis unresponsive to corticosteroid treatment. Examination revealed acuities of 20/25 in the right eye and 20/20 in the left with normal intraocular pressures. The right eye was significant for no anterior chamber cells or flare, dilated iris vessels, an IOL without deposits, 3+ vitreous cells with trace haze, and peripheral pigmentary degeneration. The left eye was normal with the exception of trace vitreous cells and a choroidal naevus. A diagnostic vitrectomy was performed in the right eye. A portion of the vitreous specimen was cultured for fungi, aerobic and anaerobic bacteria, and the remainder was processed for cytopathological examination. All cultures for micro-organisms were negative.

The vitreous supernatant and unstained cytopathological slides were sent to the National Eye Institute for further evaluation. Vitreal analysis for interleukin 2 (IL-2), IL-4, IL-6, IL-10, IFN-γ, and TNF-α using ELISA (Endogen, Woburn, MA, USA) revealed undetectable cytokine levels. The vitreous slides were stained with Giemsa, Gram, and immunohistochemical stains for T cells, B cells, and macrophages. Cytopathological examination showed clusters of macrophages admixed with CD4+ and CD8+ T cells and B cells (Fig 1A). Gram positive bacilli were seen in the cytoplasm of a few macrophages (Fig 1B). The engulfed bacilli were microdissected under a microscope with a 30 gauge needle and submitted for PCR.1 Nested PCR with P acnes specific oligodeoxynucleotide primers complementary to regions of 16S rDNA was used.1 The primers were Pa1, AAG GCC CTG CTT TTG TGG; Pa2, TCC ATC CGC AAC CGC CGA A; and rPa3, ACT CAC GCT TGG TCA CAG. Nested-PCR analysis revealed P acnes (Fig 2). A diagnosis of delayed postoperative endophthalmitis was made.

Comment

The most common causes of vitritis in elderly patients are acquired or postoperative infections, sarcoidosis, and intraocular malignancies masquerading as uveitis.1 An early diagnostic procedure is indicated if postoperative endophthalmitis is suspected. In this case, although the chronic inflammation and intracytoplasmic Gram positive bacilli in a few macrophages suggested an infectious process, the negative cultures precluded the diagnosis of an infectious endophthalmitis. To further investigate the possibility of a bacterial infection nested PCR was performed on the micro-dissected bacilli. Molecular analysis verified the presence of P acnes and a diagnosis of delayed postoperative endophthalmitis was confirmed.

Vitreous cultures are positive in less than 50% of postoperative endophthalmitis cases. In a study of 25 patients with delayed onset endophthalmitis aqueous culture and microscopy were diagnostic in 0% of cases, vitreous culture was positive in 24% and PCR from the aqueous and vitreous yielded a positive diagnosis in 84% and 92%, respectively.4 Treatment of P acnes endophthalmitis includes intravitreal vancomycin plus consideration of pars plana vitrectomy with or without capsulotomy with or without IOL removal. Although aggressive surgical intervention eradicates the infection similar visual outcomes are reported with more limited surgical treatment.

In our case the intracytoplasmic bacteria in the macrophages were the only evidence of a bacterial infection. To detect the presence of P acnes we referenced the PCR method described by Hykin that used 150 μl of the vitreous for treatment and 100 μl for PCR.5 Using the technique of microdissection and PCR with a similar volume of vitreous we additionally performed cytology and cytokine analysis which are helpful in the diagnosis of other causes of vitritis.6 This case further illustrates the benefits of molecular analysis for the diagnosis of culture.
negative delayed onset endophthalmitis. It also describes for the first time microdissection and PCR for the evaluation of endophthalmitis. Advantages of this technique are that it allows for a more comprehensive pathological examination on a limited specimen and provides the option of having the molecular studies being performed elsewhere.

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6 Buggage RR, Whitcup SM, Nussenblatt RB, et al. Using interleukin 10 to interleukin 6 ratio in the form of corneal ulcer, radiation induced conjunctival hemorrhage and local chemosis have been reported, as well as minor transient systemic effects including headaches, nausea, chills, and myalgia. Administration of intravitreal IFN-α is also a relatively simple and quick procedure. It shows great promise as a first line agent to treat conjunctival lymphoma, but long term follow up is needed.

Interferon treatment of childhood conjunctival lymphoma

Mucosa associated lymphoid tissue (MALT) lymphoma is the most common ocular adnexal neoplasms in children. These neoplastic lesions have a more indolent course than non-MALT lymphomas, and are usually found in the older age groups (<50–70 years). They are usually limited to generalised (stage 1) disease at presentation and, radiotherapy and chemotherapy have been the mainstay of treatment. 1

Case report

A 15 year old male was referred by an ophthalmologist after an 8 month history of unusual painless follicles at both nasal fornices (Fig 1A). There were no visual symp-

Figure 1 [A] Conjunctival MALT lymphoma, nasal fornix of left eye. (B) Histological section of conjunctival mucosa demonstrating dense lymphoid infiltrate (haematoxylin and eosin, original magnification ×200).

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A 15 year old male was referred by an ophthalmologist after an 8 month history of unusual painless follicles at both nasal fornices (Fig 1A). There were no visual symp-

Figure 1 [A] Conjunctival MALT lymphoma, nasal fornix of left eye. (B) Histological section of conjunctival mucosa demonstrating dense lymphoid infiltrate (haematoxylin and eosin, original magnification ×200).

Interferon treatment of childhood conjunctival lymphoma

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Figure 1 [A] Conjunctival MALT lymphoma, nasal fornix of left eye. (B) Histological section of conjunctival mucosa demonstrating dense lymphoid infiltrate (haematoxylin and eosin, original magnification ×200).
Unilateral corneal anaesthesia and ulceration following squint surgery in a child with Pendred syndrome and bilateral sixth nerve palsy

We present a 4 year old child with Pendred syndrome and bilateral sixth nerve palsy. To our knowledge this association has not been previously reported. In addition, this patient developed unilateral corneal ulceration with associated corneal anaesthesia following squint surgery. We will discuss the pathophysiology of this unusual complication following squint surgery.

Case report

This patient presented when he was 6 months old with right congenital ptosis. He was diagnosed with Pendred syndrome (sensoneural hearing loss and thyroid dysfunction) by the paediatricians and the otolaryngologists following abnormal thyroid function tests and a computed tomograph (CT) scan of the temporal bones showing Mondini malformations of both cochleae. At presentation his visual acuities were 6/60 right eye and 6/36 left eye using the Cardiff acuity cards. He had bilateral alternating esotropia with an inability to abduct either eye. There was no globe retraction or abnormal lid movements and a magnetic resonance imaging (MRI) scan had shown congenital absence of the auditory nerves but no other abnormality. A diagnosis of bilateral sixth nerve palsy was made. The squint was cosmetically poor and measured at 45 prism diptres in the distance and near. He had high hypermetropia with no significant anisometropia. Fundoscopy was normal. He was reviewed regularly in the paediatric eye clinic over the next 3 years during which time his visual acuities were within normal limits, the best recorded acuity being 6/9 right eye and 6/9 left eye using singles.

When he was 4 years old, he underwent bilateral superior rectus and inferior rectus lateral transpositions under general anaesthesia, which was uneventful with no immediate postoperative complications, and a cosmetic result (Fig 1). Alternating convergent squint surgery.

Two months later he developed a left inferior corneal ulcer (Fig 1) with surrounding punctate epitheliopathy which surprisingly did not seem to cause him much distress as expected. The left corneal sensation was definitely reduced compared to the right which appeared normal. Sensation was assessed clinically (an anaesthesiometer was not available), and was consistently reproducible by different ophthalmologists. There was no exophthalmos or any other sign of thyroid orbitopathy. The right eye remained asymptomatic. Empirical therapy with topical ofloxacin and lubricants was unhelpful. He proceeded to have glue tarsorrhaphy which transiently aided the healing of the corneal ulcer. However, the ulcer quickly recurred when the tarsorrhaphy reversed. He subsequently had left inferior lid shortening with a canthal sling to elevate the lower lid to protect the corneal epithelium. The ulcer resolved leaving an area of corneal scarring. He is being reviewed regularly in the eye clinic.

Comment

Pendred syndrome is an autosomal recessive disorder characterised by congenital deafness and thyroid goitre. The hearing loss is usually severe and is present at birth, and the goitre generally appears at puberty or later but may be present in early childhood with an associated euthyroid or hypothyroid state. Affected individuals are reported to be otherwise normal.

The pathophysiology of the corneal anaesthesia and ulceration in this patient is uncertain. There are several contributory reasons for the corneal anaesthesia. They include herpes simplex keratitis, postoperative anterior segment ischaemia, surgical trauma to the long posterior ciliary nerves or ciliary ganglion, congenital absence of sensation, and surgery reducing Bell's phenomenon.

The clinical course was not typical of herpes simplex and there was no previous history of corneal pathology. Postoperative anterior ischaemic syndrome was unlikely as only two recti muscles were operated on and no anterior uveitis was observed. To our knowledge there are no reported cases of corneal anaesthesia after squint surgery. There was no evidence of pre-existing involvement, which one may expect with trauma to the long posterior ciliary nerves or ciliary ganglion.

Congenital absence of corneal sensation was the most likely cause, especially in view of his unusual cranial nerve anomalies, and we believe he had pre-existing corneal anaesthesia before squint surgery despite the absence of any other fifth cranial nerve signs. Following the lateral transposition of the superior rectus his Bell's phenomenon was noted to be absent thereby compromising his corneal protection. In addition, he was observed to have significant lagophthalmos while asleep. We believe that the combination of corneal anaesthesia, abolished Bell's phenomenon, and lagophthalmos compromised his corneal integrity resulting in corneal ulceration.

This case highlights the importance of determining corneal sensation before transposition surgery on the superior rectus as Bell's phenomenon may be abolished therefore compromising corneal protection. This is especially relevant in patients with unusual cranial neuropathy and lagophthalmos.

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Gemella haemolysans acute postoperative endophthalmitis

Endophthalmitis is perhaps the most feared complication of cataract surgery, with a reported incidence between 0.01% and 0.13%.

The most common organisms reported in the literature, which identified Gemella haemolysans from both anterior chamber and vitreous aspirates. The organism was reported to be sensitive to gentamicin, ciprofloxacin, laevofloxacin, amoxicillin/clavulanate, chloramphenicol, and resistant to trimethoprim.
The patient continued to make steady progress; 2 months later vision had improved to 6/9 unaided. The patient at that time was progressing; 2 months later vision had improved.

**Comment**

Gemella haemolysans is an aerobic or facultative anaerobic, Gram positive coccus, a normal commensal of the oral cavity and upper respiratory tract of low virulence. Systemic infection may lead to septic shock, meningitis, arthritis, or pneumonia, all of which are rare. Identification is difficult. Though Gram positive, the cocci are easily decolorised and hence may appear Gram variable or even negative.

Initially Gemella was included under the genus Neisseria but is now classified as a separate genus within the family Streptococcaceae. No studies on susceptibility to antiseptics have been published, though there is no reason to believe that it may be resistant to povidone-iodine preparations. The organism is sensitive in vitro to penicillin, streptomycin, vancomycin, chloramphenicol, and tetrasulphathiazole.

A literature search revealed only one previously reported case of infection by Gemella haemolysans, with ketorolac and consecutive endophthalmitis. Interestingly this patient was reported to have active sarcoidosis on admission. This possible association between sarcoidosis and infection by Gemella may be purely coincidental, as no such association has been reported with systemic infection.

Gemella haemolysans is difficult to identify, because of its close resemblance to viridans streptococcus and Neisseria. As diagnostic technology improves, Gemella haemolysans endophthalmitis may be described more often in the future. This report highlights the importance of infection with rare commensal organisms in healthy, immunocompetent individuals after uneventful phacoemulsification cataract surgery.

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**Does topical brimonidine tartrate help NAION?**

There is no proved treatment for non-arteritic anterior ischaemic optic neuropathy (NAION). Topical brimonidine tartrate has been reported to have a neuroprotective benefit for retinal ganglion cells following experimental elevation of intraocular pressure and optic nerve injury in the rat, which is blocked with coadministration of the α2 antagonist, rauwolscine. Increased retinal ganglion cell survival has also been shown to occur following oral administration of brimonidine in monkeys with experimental glaucoma. These results were the basis of the recently aborted clinical trial of topical brimonidine purite for acute NAION and our retrospective study of 31 patients with NAION, who were evaluated within 3 weeks of the onset of visual loss and were followed up for a minimum of 8 weeks. During 2001–2, we treated all (14) patients with brimonidine tartrate within 14 days (mean 3.5, SD 5.52) of the onset of visual loss. Five patients were treated after 1 day of symptoms. Three were treated within 1 day of the onset (mean 24 h, SD 0.4), eight were treated within 2 days of the onset (mean 1.5, SD 1), and three were treated within 3 days of the onset (mean 2.1, SD 0.5). The visual acuity was 0.29 (SD 0.70; p=0.04) for untreated patients. The mean colour vision was 0.42 (SD 0.41) for treated and 0.35 (SD 0.46; p=0.43) for untreated patients.

For the masked examiner's evaluation, the mean baseline visual field (2.0, SD 0.91) was similar to the field (1.93, SD 0.96; p=0.85) for controls. At the 8–12 week examination, the mean visual field grade was 2.2 (SD 0.81) for treated and 1.0 (SD 0.70; p=0.04) for untreated patients. The mean colour vision was 0.42 (SD 0.41) for treated and 0.35 (SD 0.46; p=0.43) for untreated patients.

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is possible earlier treatment might have been more effective, although patients who worsened received treatment sooner than those who did not worsen. Increased dosing frequency or using a different preparation of bromidrine might be more effective. Additionally, the number of subjects in the study was small and a negative trend could appear more profound.

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References


Chronic eye movement induced pain and a possible role for its treatment with botulinum toxin

Chronic ocular pain may have many causes and can be a frustrating problem for both patient and doctor alike. We describe two patients who had similar symptoms and eye findings who had been unable to relieve their pain with conventional anaesthetics. We postulate a cause for their pain and describe our experience of a treatment strategy using a standard dose of botulinum toxin injection into an extraocular muscle.

Case 1

A 56 year old white woman presented with a history of bilateral ocular pain and a possible role for its treatment with botulinum toxin. She had a 9 month course of oral steroids and despite this needed tramadol, paracetamol, and flurbiprofen to control her pain. Her symptoms and examination findings slowly stabilised until she was left with marked limitation of upgaze in her right eye. Her symptoms did not change over the next 3 years, at which point she was referred to our care. When she attempted to look up she described a juddering sensation and severe pain just above the eye. She rarely had pain at night but was still using regular oral ibuprofen for pain relief. Her pain was exacerbated by reading or looking at the computer and she complained of vertical diplopia.

On examination she had limitation of abduction and elevation of her right eye and prisms did not improve her symptoms. A tentative diagnosis of inflammatory spasm was made. She was treated with botulinum toxin injection to her right inferior rectus. Two weeks later there was much less tightness and discomfort in the orbit but she had diplopia in all positions of gaze and was forced to occlude one eye. Three months later the pain was much improved but she realised she had found the diplopia intolerable and declined further treatment.

Case 2

A 46 year old white man presented complaining of chronic constant ocular discomfort which followed strabismus surgery 8 years earlier for an A-pattern exotropia with diplopia on downgaze. The pain was worsened by prolonged television watching and prisms in his glasses did not help. Pain was much worse on upgaze and right gaze, which were limited. Oral non-steroidal anti-inflammatory agents (NSAIDs) did reduce the pain a little but only when taken in high doses (100 mg three times daily flurbiprofen).

On examination he had a right hyperphoria, with an A-pattern exotropia and an abnormal head posture for distance. He still had diplopia. Botulinum toxin was injected into his left medial rectus muscle, which resulted in a profound reduction in his symptoms, leaving him with a small exophoria. His diplopia resolved completely after 10 weeks.

The “pressure sensation” and pain in the right eye recurred after about 6 months, this time with no diplopia. He had a further injection of toxin 8 months after the first which again significantly improved his pain but gave him diplopia for 3 weeks. He continues to take flurbiprofen 50 mg three times daily orally.

Comment

The pain demonstrated by these two patients is typically much worse in certain directions of gaze and particularly during prolonged gaze holding such as when reading or watching television. It had a clear precipitating event and the most remarkable feature is that it had persisted for over 2 years in each case without significant head or eye movement or regression. No active disease process could be found to account for the continued pain. The pain is severe and responds only to high doses of analgesics, particularly NSAIDs. None of our patients felt that their pain was satisfactorily controlled by their analgesics.

We believe that there may be a process of chronic low grade inflammation affecting the extraocular muscles and the tissues around them which is exacerbated by continued contraction and relaxation of the same muscles. Muscular spasm perhaps triggered by this inflammatory process may be the cause of the most severe pain and this could account for the exacerbations of pain in certain directions of gaze and on prolonged gaze holding activities. Ocular muscle ischemia, perhaps caused by constricting scar tissue, remains a possibility but the onset of the pain is very fast making this less likely.

The pain relief seen in our patients may simply be the result of paralysing an inflamed muscle but there is growing evidence for a separate antinociceptive effect of botulinum toxin. No direct peripheral cutaneous antinociceptive effect could be shown by Biersch et al., however inhibition of release of substance P has been demonstrated in vitro and it can be hypothesised that botulinum toxin treatment may reduce the local release of nociceptive neuropeptides from either cholinergic neurons or from C or A delta fibres in vivo. The mechanisms by which botulinum toxin may relieve pain, including a possible analgesic effect of botulinum toxin metabolites, are reviewed by Guyer.

There is a growing literature on the use of botulinum for painful conditions, particularly those in which muscle spasm plays a part. These include writer’s cramp, postoperative pain in spinal cerebral palsy, and perhaps more surprisingly perhaps tic douloureux and painful tic convulsif. Many of the reported uses are single case studies and not all controlled trials have shown a positive effect of treatment.

It is not possible to rule out a powerful placebo effect in our patients but, whatever the mechanism of action, their pain was vastly improved and botulinum toxin treatment is very safe in competent hands.

In the cases described both botulinum toxin served a dual purpose in that it had the potential to improve their ocular deviation for which it is well known and it also reduced the severe ocular discomfort. Unfortunately, the resulting diplopia limited its usefulness in one case but we feel that this treatment should be considered in this unusual group of patients who present a difficult management problem even to the most experienced ophthalmologists.

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References


Intrastromal lamellar femtosecond laser keratoplasty with superficial flap

Lamellar keratoplasty has usually been performed taking a trephine to delineate the extent of the tissue to be excised, and a knife or similar instrument to remove the lamellar corneal tissue from the underlying deep corneal bed. In a similar way, the lamellar donor tissue was prepared and inserted into the recipient bed. The depth of the lamellar excision extended from the corneal epithelial surface to the deep corneal stroma. Marked disadvantages of the technique have been pronounced corneal astigmatism and optical insufficiencies of the interface between the lamellar graft and the recipient corneal bed caused by irregularities of both surfaces. The purpose of the present report was to describe the new femtosecond laser technology, which may enable us to perform a new type of intrastromal lamellar keratoplasty with preservation of an intact Bowman’s layer and regular corneal epithelium.

Case report

Using a corneal contact lens and a femtosecond laser (20/10 Perfect Vision, Am Taubenfeld 21/1, D-69123 Heidelberg, Germany) with a wavelength of 1060 nm, a spot size of about 10 μm, and a laser pulse duration of several hundred femtoseconds, a pre-descemetal incision running parallel to the corneal surface was created in five postmortem eyes of slaughterhouse pigs. The diameter of the deep stromal incision was 7 mm. In a second step, a circular sagittal incision was performed starting from the peripheral edge of the already existing incision in the pre-descemetal level to the superficial layer of the central cornea. In continuation of the latter sagittal incision the corneal flap was prepared with a diameter of 7 mm, a thickness of about 100 μm, a hinge, and three positional pikes. The pikes in the flap with the corresponding notches in the bed of the flap were formed to increase the rotational stability of the flap after repositioning. The height of the peaks was about 0.40 mm. After opening of the flap the intrastromal segment situated between the pre-descemetal incision and the incision in the superficial stromal level was removed and exchanged against a similar formed segment obtained from another (donor) pig eye, and the flap was repositioned.

For all eyes included in the study, the intrastromal corneal button and the superficial flap with the three positional pikes could be prepared without major difficulties. The corneal buttons could easily be repositioned into their original beds as well as into the recipient beds of other eyes in which the recipient pikes were created with the same diameter as the donor button. The time taken for preparation of the intrastromal corneal button and the corneal flap, and for the exchange of the corneal buttons was less than 10 minutes in all cases.

Comment

Femtosecond laser technology allows a new type of intrastromal lamellar keratoplasty with removal of a mid-stromal segment and preservation of an intact Bowman’s membrane. Considering the decreased amount of allogenic corneal tissue transplanted, and regarding the preservation of the original corneal surface, lamellar intrastromal femtosecond laser keratoplasty may be associated with a smaller rate of immunological graft reaction and with a lower postoperative corneal astigmatism in some eyes. Future clinical studies may show whether positional edges in the superficial flap increase its postoperative rotational stability.

Proprietary interest: none

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References


Demographic study of paediatric allergic conjunctivitis within a multiethnic patient population

From October 1999, all patients referred to the paediatric ophthalmology service in Bradford have been added to a computerised database. This is the only paediatric ophthalmology service in Bradford. All patients referred to the paediatric ophthalmology service within the city of Bradford and receives all GP referrals of this type. Patients with a clinical diagnosis of chronic allergic conjunctivitis were identified from October 1999 to July 2001. There were 140 patients fulfilling these criteria. The relative prevalence of chronic conjunctivitis in the community is discussed.

Clinical data

Forty three patients were identified from the database; 39 patients fulfilled entry criteria for this study and records were retrieved for 35. There were 24 Asians and 11 white children.

For Asian patients, the mean age was 9.58 (SD 4.02) years. For the white patients, the mean age was 7.82 (SD 3.19) years. Follow up ranged from 3–14 months, mean 6 months. The prevalence of allergic conjunctivitis in Asians was 59 per 100 000 (24 in 40 524) and in white children, 12 per 100 000 (11 in 93 988); a relative prevalence of 5 to 1 (χ² test p < 0.001).

There was a predominance of males in both ethnic groups, 2.4:1 in Asians and 1.8:1 in white children. This difference in sex was not significant between both groups (Fisher’s test, p = 0.71).

The overall age distribution for all males was 4.54 years and for all females was 10.01 years. For Asians, the mean age for males was 9.18 years and for females was 10.57 years. For white children, the mean age for males was 7.00 years and for females was 9.25 years.

Corneal complications

There were 14 with punctate epithelial erosions (10 Asians and four white children). Comparing patients from both groups with severe disease, there was a relative prevalence of Asians by 6.75 to 1 (Fisher’s test, p = 0.001). In two cases, visual loss occurred after the onset of chronic conjunctivitis due to corneal plaque and corneal pannus. Both were Asian.

Comment

Various studies have reported allergic eye disease to be more common among Asian and black patients. This may be due to genetic and environmental factors. We found allergic eye disease to be more common in Asians than white children. It is possible that ocular allergy is multifactorial but perhaps with a greater genetic predisposition in certain ethnic communities. We could not comment on the prevalence of chronic allergic conjunctivitis in the community because of referral bias since we only see patients referred by GPs. The extent to which milder cases are treated in the community is not known but we feel that the more severe cases are the ones referred to our department. Our findings highlight that allergic eye disease appears to be more common and complicated in Asian patients in the Bradford population. This potential risk of sight threatening disease means that they are more likely to require topical steroid treatment. This has led us to recognise that appropriately aggressive treatment is essential in these patients.

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References

Surgical Eye Expeditions International
Volunteer ophthalmologists in active surgical practice are needed to participate in short term, sight restoring eye surgery clinics around the world.
Contact: Harry S Brown, Surgical Eye Expeditions International, 27 East De La Guerra, C-2, Santa Barbara, CA 93101–9588, USA (tel: +805 963 3303; fax: +805 963 3564; email: hsbrown.md@cox.net or seeinl@seeinl.org; website: www.seeinl.org).


ing in organ transplant numbers
According to UK Transplant, the UK has seen the highest number of organ transplants in six years, from 30 March 2001 to 31 March 2003) 2777 patients had their lives saved or dramatically improved through the generosity of 1064 donors. This equated to a 6% increase compared to the previous 12 months (1 April 2001 to 31 March 2002). Furthermore during 2002–3, the highest number of people benefited from a corneal transplant for five years (1997–98) and 240 more people had their sight restored than the previous year. For further information see UK Transplant’s website (www.uktransplant.org.uk).


current international organisation on: Retinal and Vitreous Surgery with Case Presentations preceding the Annual Meeting of Iranian Society of Ophthalmology
The detachment course with international faculty on: Retinal and Vitreous Surgery with Case Presentations preceding Annual Meeting of Iranian Society of Ophthalmology will be held


Glaucome Society 24th Annual Meeting and Dinner
The Glaucome Society 24th Annual Meeting and Dinner will take place on 20 November 2003, from 8.30 am to 11.30 pm at the College of Physicians, London, UK. Further details: Ms Janet Flowers (email: glaucoc@ucl.ac.uk; website: www.glaucome.org).
5th International Symposium on Ocular Pharmacology and Therapeutics (ISOPT)

The 5th International Symposium on Ocular Pharmacology and Therapeutics (ISOPT) will take place 11–14 March 2004, in Monte Carlo, Monaco. Please visit our website for details of the scientific programme, registration, and accommodation. To receive a copy of the Call for Abstracts and registration brochure please submit your full mailing details to http://www.kenes.com/isopt/interest.htm.


XVth Meeting of the International Neuro-Ophthalmology Society

The XVth Meeting of the International Neuro-Ophthalmology Society will take place 18–22 July 2004, in Geneva, Switzerland.

Further details: Prof. A Safran, University Hospital Geneva, c/o SYMPORG SA, Geneva (fax: +41 22 839 8484; email: info@symporg.ch; website: www.symporg.ch).