Endophthalmitis Following Strabismus Surgery: IPOSC Global Study

Ofira Zloto\textsuperscript{ab}, Eedy Mezer\textsuperscript{cd}, Luis Ospina\textsuperscript{e,f}, Branislav Stankovic\textsuperscript{gh}, and Tamara Wygnanski-Jaffe\textsuperscript{ab}

\textsuperscript{a}Goldschleger Eye Institute, Sheba Medical Center, Tel Hashomer, Israel; \textsuperscript{b}Sackler Faculty of Medicine, Tel-Aviv University, Tel Aviv, Israel; \textsuperscript{c}Department of Ophthalmology, Ruth Rappaport Children’s Hospital, Rambam Health Care Campus, Haifa, Israel; \textsuperscript{d}Bruce Rappaport Faculty of Medicine, Technion, Israel Institute of Technology, I.I.T., Haifa, Israel; \textsuperscript{e}Ophthalmology Department, Sainte-Justine’s Hospital, University of Montreal, Montreal, Canada; \textsuperscript{f}Ophthalmology Department, Notre-Dame Hospital, University of Montreal, Montreal, Canada; \textsuperscript{g}Faculty of Medicine, University of Belgrade, Serbia; \textsuperscript{h}Institute of Ophthalmology, Clinical Centre of Serbia, Belgrade, Serbia

\textbf{ABSTRACT}

\textbf{Purpose:} To examine the characteristics of patients with endophthalmitis after strabismus surgery (PSSE), the characteristics of the strabismus surgery, treatment, and prognosis as reported by pediatric ophthalmologists who are members of the American Association for Pediatric Ophthalmology and Strabismus (AAOPS) around the world.

\textbf{Methods:} An email communication was sent to all members of AAPOS. The email included a web link to a survey that included 34 questions that examined the characteristics of patients with endophthalmitis after strabismus surgery.

\textbf{Results:} Three hundred forty-nine ophthalmologists responded to the questionnaire. Two hundred sixty four (76\%) had practiced pediatric ophthalmology for over 10 years. Twenty-seven cases of endophthalmitis were reported. The medial rectus muscle (MR) was the most common muscle undergoing surgery (55\%). In 5 patients (21\%), perforation was observed at the time of surgery. In all cases, endophthalmitis was diagnosed 3–7 days after surgery. The final visual acuity of 34\% was no light perception (NLP).

\textbf{Discussion:} This series is the largest existing series on PSSE. Our data suggest that PSSE can occur also in healthy patients and in patients without a visualized scleral perforation. The majority of PSSE occurs during the first week after surgery. Therefore, we believe that all patients that underwent strabismus surgery should be examined in this time frame. The prognosis was poor with 48\% demonstrating a visual acuity of LP or NLP at the final follow-up.

\section*{Introduction}

Post strabismus surgery endophthalmitis (PSSE) is one of the most devastating complications of eye surgery resulting possibly in blindness and even loss of the eyeball.\textsuperscript{3} It is considered a very rare complication of strabismus surgery with an incidence of 1/30,000 to 1/185,000 cases.\textsuperscript{2} Even in cases in which a scleral perforation has been encountered, endophthalmitis is still very rare, occurring in only 0.4\%.\textsuperscript{3} Other postulated mechanisms for postoperative endophthalmitis include endogenous seeding and intraocular progression from a localized abscess of the sclera.\textsuperscript{4} Various pathogens have been found in different studies, including: \textit{Staphylococcus aureus}, \textit{Haemophilus influenzae}, \textit{Pseudomonas aeruginosa}, \textit{Staphylococcus epidermidis}, and \textit{Staphylococcus coagulase}.\textsuperscript{5,6} The visual prognosis in most cases is poor.\textsuperscript{6,7}

Most of the literature on PSSE includes case reports and small case series.\textsuperscript{5,8,9} Those case series are summarized in Table 1. The largest series, including 22 patients, was presented recently by Simonsz in the World Society of Pediatric Ophthalmology and Strabismus (WSPOS) Subspecialty Day.\textsuperscript{17}

The purpose of the current study was to examine the epidemiological and general health characteristics of patients with PSSE after strabismus surgery, the characteristics of the strabismus surgery, as well as clinical findings, treatment, and prognosis as reported by pediatric ophthalmologists who are members of the American Association for Pediatric Ophthalmology and Strabismus (AAPOS) around the world, with the goal of attaining a larger series from which one could possibly acquire more knowledge on PSSE and perhaps draw practical ways to decrease this postoperative complication.

\section*{Methods}

\textbf{Survey population and questionnaire}

An email communication was sent to all members of AAPOS through the International Pediatric Ophthalmology and Strabismus Council (IPOSC) secretariat, between May and July 2016. The email included a web link to a survey (created on \url{www.surveymonkey.com}), explaining the purpose of the study and offering a contact email and phone number that allowed the respondents to seek further clarification if needed. Follow-up emails were sent in August 2016 to those who had not yet responded. The questionnaire included (Supplementary Methods) 34 questions that examined the characteristics of patients (adults and children) with endophthalmitis following strabismus surgery in all years of practice of the strabismus'
surgeon, the type of surgery and technique utilized, clinical findings, treatment, and outcome of PSSE in different countries around the world.

All authors declared not to have financial or non-financial conflict of interest, and no formal consents were required given the retrospective nature of the study.

Statistical analysis

Statistical analysis, including distribution, was performed with JMP Statistical Discovery Software 7.0 (SAS Institute, Cary, NC, USA). The overall significance level was set to an alpha of 0.05.

Results

Three hundred forty-nine AAPOS member ophthalmologists (out of 449 ophthalmologists that are listed on email lists, 78% response rate) who were members of AAPOS responded to the questionnaire. Forty-nine (14%) had practiced pediatric ophthalmology for less than 5 years, 36 (10%) for 5–10 years, and 264 (76%) for over 10 years. Eighty-two ophthalmologists (23%) preformed up to 50 surgeries per year, 95 (26%) preformed 50–100 surgeries per year, and 177 (51%) preformed over 100 strabismus surgeries per year.

Three hundred twenty-one (92.5%) were right handed and 26 (7.5%) left handed.

The geographical distribution of practice around the world is summarized in Figure 1.

Incidence and characteristics of patients

Twenty-seven cases of endophthalmitis were reported. Eighteen cases (72%) were seen and operated by the ophthalmologists responding the survey and seven (28%) were referred to them by a different surgeon. Four (15%) had a medical history of central nervous system disease prior to surgery, four (15%) had a concurrent respiratory tract infection, one (4%) had a history of immunosuppression, and one (4%) had a history of nasolacrimal duct obstruction or infection. Sixteen (62%) were healthy, with no concurrent or past medical history, and the health status of one patient was not known. There were no reported cases of PSSE in highly myopic patients. The distribution of the ages of the patients is summarized in Figure 2.

Table 1. Case series of post-strabismus surgery endophthalmitis.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Number of cases</th>
<th>Post medical history</th>
<th>Muscle involved in strabismus surgery</th>
<th>Was a perforation found</th>
<th>Prognosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Simon JW</td>
<td>1992</td>
<td>3</td>
<td>None</td>
<td>Not written</td>
<td>3 cases (100%)</td>
<td>1- Impaired vision (33.3%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>2- NLP (66.67%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>3- Enucleation (50%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>4- NLP (100%)</td>
</tr>
<tr>
<td>Recchia FM</td>
<td>2000</td>
<td>6</td>
<td>3 (50%)</td>
<td>SR resection (16.67%)</td>
<td>None</td>
<td>5- NLP (100%)</td>
</tr>
<tr>
<td>Huang S</td>
<td>2011</td>
<td>2</td>
<td>None</td>
<td>MR resection (83.33%)</td>
<td>2 cases (100%)</td>
<td>6- Enucleation (50%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>7- Enucleation (50%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>8- NLP (100%)</td>
</tr>
<tr>
<td>Alniemi ST</td>
<td>2016</td>
<td>3</td>
<td>2 (66.67%)</td>
<td>LR resection (33.33%)</td>
<td>None</td>
<td>9- NLP (100%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>10- Enucleation (50%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>11- NLP (100%)</td>
</tr>
<tr>
<td>SimontzHJ</td>
<td>2016</td>
<td>24</td>
<td>18 (75%)</td>
<td>LR resection (29.16%)</td>
<td>4 cases (16.67%)</td>
<td>12- NLP (100%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>IR resection (4.16%)</td>
<td></td>
<td>13- Enucleation (50%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>MR resection (66.67%)</td>
<td></td>
<td>14- NLP (100%)</td>
</tr>
</tbody>
</table>

Figure 1. The geographical distribution of participants' practice.

Figure 2. The distribution of ages of patients with post-strabismus surgery endophthalmitis.
Strabismus surgery

In 11 eyes (61%), the surgery was performed on the right eye, and in seven eyes (39%) on the left eye. The medial rectus muscle (MR) was the most common muscle undergoing surgery (55%), followed by the lateral rectus (LR) (15%) and the inferior rectus (IR) (14%).

The distribution of muscles involved in PSSE is summarized in Figure 3.

Twenty-four muscles (89%) were sutured directly to the sclera, and three muscles (11%) were hanged back from the original insertion (1 IR, 1 LR, 1 MR). Ten surgeons (44%) used a Vicryl absorbable suture, 6-0, with a double-armed S29 needle, six (26%) used Vicryl absorbable suture, 6-0, with a double-armed S24 needle, four (17%) used Vicryl absorbable Suture, 6-0, with a double-armed S14 needle, and 3 (13%) used different sutures.

Seven (29%) used Tegaderm or other methods to cover the eyelids or eyelashes during surgery, while 17 (71%) did not report they routinely cover the eyelids or eyelashes.

Nineteen (83%) sutured the conjunctiva, four (17%) did not, and four did not answer the question.

In five patients (21%), a perforation was observed at the time of surgery. No one reported diagnosing a perforation after the surgery. In two patients, the perforation was treated by cryotherapy, in one by laser, and in one by sutures.

Twenty-two patients received antibiotics after the surgery by different routes as summarized in Table 2.

Characteristics of endophthalmitis

The first most common ocular signs of PSSE included: pain in nine (38%) patients, extraocular signs of inflammation such as eyelid edema, proptosis, and restricted ocular motility in seven (29%) patients, conjunctival signs such as conjunctival injection and chemosis in three (13%) patients, and vitreous involvement: vitreitis and snow balls in two (8%) patients. Other initial presentations included: decreased visual acuity (VA) or symptoms that motivated seeking care at emergency services. The latter delayed the diagnosis until referral to ophthalmic care centers in three (12%) patients.

The first systemic clinical signs of PSSE were: fever in six (35%) patients, lethargy in six (35%) patients, headache in four (24%) patients, and sepsis in one (6%) patient.

In all cases, endophthalmitis was diagnosed 3–7 days after surgery.

The distribution of visual acuity at presentation is summarized in Figure 4.

In 14 patients, the diagnosis was made by pars-plana vitrectomy, five patients underwent a vitreous tap and an...
injection of antibiotics to the vitreous cavity, and nine patients underwent a steroid and antibiotics injection to the vitreous cavity. Twelve patients (52%) underwent an anterior chamber tap and injection of antibiotics. In one patient, the treatment was not specified, and data could not be recovered.

**Prognosis**

The final VA after PSSE was no light perception (NLP) in 34%, 14% light perception (LP), and 12% hand motion (HM). The distribution of final visual acuity is summarized in Figure 5. Two eyes underwent enucleation or evisceration.

**Discussion**

The literature on PSSE is sparse, and prospective studies on the subject are unlikely to be undertaken in the near future. Little has been documented concerning the characteristics and treatment of this rare but devastating complication. This survey summarizes the clinical presentation, diagnosis, characteristics of surgery, and treatment and outcome of patients with PSSE from around the world. There is large representation from North America, Europe, and the Middle East and a smaller number of reports from Africa, Australia, the Far East, and South America, which may reflect a lower number of strabismus surgeons practicing in these regions and the lower number of physicians who are member of the AAPOS.

Most of the ophthalmologists replying to the questionnaire had over 10 years of clinical practice in pediatric ophthalmology and a surgical volume of more than 100 surgeries per year. This may suggest that inexperience is not a substantial factor associated to PSSE.

Simonsz et al. gave an oral presentation in the WSPOS Subspecialty Day in 2016 and among 22 cases of PSSE. They concluded that PSSE occurs most often after medial rectus recession in either very young or very old patients and that it was diagnosed 2–8 days after surgery. Recchia et al. published the largest series in the literature. Their series included six cases of PSSE, reporting that children may not recognize or verbalize symptoms. However, lethargy, asymmetric eye redness, eyelid swelling, or fever in the postoperative period should prompt urgent ocular examination. The diagnosis of PSSE may be made when slit lamp or indirect ophthalmoscopic examination reveal vitreous opacification with or without a hypopyon. To date, these two reports were the largest series in the literature. In this survey, we reported 27 cases, which make this series the largest reported to date.

There is no conclusive evidence to which mechanisms cause PSSE. Whether a history of respiratory or lacrimal infection or an immunosuppression status is predisposing factors remains unknown.

In our study, only 38% patients had prior medical history of central nervous system disease, concurrent respiratory tract infection, immunosuppression, and nasolacrimal duct obstruction or infection. In Recchia et al. series, 50% of the patients had past medical history of trisomy 21, asthma, and respiratory tract infection, while in Alniemiet et al., 66% of cases had a past medical history of trisomy 21 and colon cancer. The inconsistent findings among different studies make it difficult to draw conclusions but also indicate that healthy patients may also develop PSSE.
Endophthalmitis after globe perforation in strabismus surgery is very rare. In this study, a perforation was visualized in only 21% of the cases. This is in accordance to previous studies that also reported low rates of PSSE after globe perforation. However, it is possible that a micro perforation occurred but was not noted. Therefore, whether a perforation is necessary for PSSE to occur is still unknown.

When a perforation has occurred, most of the patients were treated with antibiotics via different routes such as intravenous or intravitreal in order to prevent PSSE. Probably this prophylactic treatment helps prevention of PSSE in some cases, because this complication occurred only in a small number of patients noted to have a perforation.

In general, the largest group of patients undergoing strabismus surgery is below 6 years old. Simonsz et al. reported in ARVO 2009 and in WSPOS 2016 that two age groups are prone to develop PSSE, which include: children under 6 years of age and elderly patients over the age of 65 years. Accordingly, they recommended postponing strabismus surgery after the age of 6 years, when possible. In our study, PSSE occurred most frequently in the 2-4-year-old age group of children and after the age of 14 years. Those undergoing surgery under the age of 6 years represented 57% of cases.

The most commonly involved muscle in PSSE in our series is MR (55% of the cases). This may reflect the fact that the MR is the most frequently operated muscle and that surgery to correct esotropia is also the most commonly performed. In addition, this may be caused by the eye structure of most patients (Caucasian). In Asia, data may be different. Simonsz and Recchia et al. also found PSSE more commonly in patients that underwent MR recession.

In our study, 71% of the surgeons did not cover the eyelid or eyelashes during strabismus surgery. Covering these areas, as performed in cataract surgery, may prevent contamination of the eye during surgery, although it was never scientifically examined in strabismus surgery in the past. Only 11% of surgeons used the hang-back technique, which may hypothetically reduce the risk for PSSE following scleral perforation, because needles are passed through thicker sclera at the original insertion.

All cases of PSSE in our study presented 4 weeks after strabismus surgery. Therefore, it may be recommended to perform during the first week after strabismus surgery a red reflex testing and a slit lamp examination of the anterior as well as a dilated fundus examination.

The endophthalmitis vitrectomy study performed on cataract surgery patients concluded that vitrectomy is not necessary for patients with a visual acuity that was better than light perception (LP). However, the results of the same study might be relevant to other types of ocular surgery. In our study, 59% of patients had an initial visual acuity better than LP. Of those 11 patients (61.11%) underwent vitrectomy. Therefore, most of the surgeons in this study did not adhere to the recommendation of the endophthalmitis vitrectomy study. The final VA of 6 (33.3%) of those patients was NLP. Therefore, in suspected PSSE cases, vitrectomy might be beneficial even for patients with VA better than LP.

The visual prognosis of PSSE was poor: 48% deteriorated to LP or no NLP at the final follow-up visit, and two patients underwent evisceration or enucleation. The visual outcome was even poorer in children with PSSE under 6 years of age, whose vision deteriorated to LP or NLP in 53.33% of the cases.

In conclusion, to the best of our knowledge, this study represents the largest series reporting on the characteristics of PSSE. We found that PSSE can also occur in healthy patients and in the absence of noticed scleral perforation. Most cases of PSSE occur within the first postoperative week, and some patients may be asymptomatic. Therefore, we believe that all patients that underwent strabismus surgery should be examined in this time frame. Children under 6 years of age may be at a higher risk of developing PSSE, but we cannot exclude over-representation of the same age group in our series. The vision prognosis of PSSE is poor. Most patients presented with visual acuity better than LP, but in 48% of patients’ vision deteriorated to worse than LP at last reported follow-up. The data we have presented from our case series was not intended to replace larger further studies that will examine sterilization techniques prior the surgery and surgical techniques and their association with endophthalmitis.

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Declaration Of Interest

The authors have no conflict of interest.

References


