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Socioeconomic and psychological impact of treatment for unilateral intraocular retinoblastoma

L’impact socioéconomique et psychologique de traitement pour le rétinoblastome unilatéral intraoculaire

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KEYWORDS
Unilateral; Retinoblastoma; Enucleation; Socioeconomic; Cancer; Developing country

Summary
Purpose. — To identify the socioeconomic and psychosocial impacts of clinical treatment decisions for advanced unilateral intraocular retinoblastoma.
Design. — Retrospective observational case series.
Methods. — Setting: institutional study at Alexandria Main University Hospital. Study population: records of 66 unilateral retinoblastoma cases treated from May 2005 to May 2013 were retrospectively reviewed. Sixty cases were eligible (International Intraocular Retinoblastoma Classification [IIRC] group C, D or E). Procedures: two treatment groups were compared:

Keywords
Unilateral; Retinoblastoma; Enucleation; Socioeconomic; Cancer; Developing country
Socioeconomic impact for unilateral RB

Introduction
Retinoblastoma (RB); although relatively rare; is the most common malignant intraocular tumor in children [1] with approximately 8000 cases/year worldwide [2]. In 2/3 of cases, RB is unilateral [3] and as the other eye is free; the diagnosis is mostly delayed till an advanced state especially in less developed countries [4]. With the current management protocols, survival is reaching towards 100% in developed countries and is improving in less developed countries [5].

The optimal management of advanced unilateral intraocular retinoblastoma (International Intraocular Retinoblastoma Classification [IIRC] groups D and E eyes, sometimes

enucleation vs. salvage treatment. Salvage treatment eyes were further subdivided based on IIRC group. Six socioeconomic parameters (financial burden, financial impact, psychological, social, medical and tumor impacts) were scored. Parameter scores ranged from 0 to 3, for overall score range 0 (no adverse impact) to 18 (severe adverse impact). Main outcome measures: derived Socioeconomic scores were correlated with treatment and outcomes.

Results. — The enucleation group (28 eyes) had a median overall Socioeconomic score of 4/18, significantly lower than the salvage treatment group (32 eyes), median score 11/18 (P < 0.01). Socioeconomic score varied with IIRC group. Attempted eye salvage failed in 25 children, due to uncontrolled tumor (44%) and socioeconomic impact of cumulative therapies (56%). Treatment duration and Socioeconomic score were higher for the 5 children in the salvage treatment group who developed metastatic disease compared to those without metastasis (P < 0.01).

Conclusions. — The socioeconomic and psychosocial impacts of attempted ocular salvage for unilateral intraocular retinoblastoma are severe, in comparison to primary enucleation. Primary enucleation is a good treatment for unilateral retinoblastoma.

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group C eyes) [6] is the subject of ongoing international debate. Standard of treatment for unilateral disease has generally been primary enucleation followed by adjuvant chemotherapy when high-risk histopathological features are observed [3,7,8]. Recently, primary attempt for ocular salvage using systemic chemoreduction with focal therapy, intra-arterial chemotherapy (IAC), or radiation therapy, has become frequent, with variable success [9,10].

In developing countries, the high costs and sometimes unavailability of, or limited training in, eye salvage treatment modalities, and the immaturity of health insurance systems, make attempt at eye salvage a tantalizing, hopeful, but difficult option for the child, the family and the treating physician [4,11,12]. Enucleation carries social stigma in some cultures, and physicians may experience difficulty in convincing parents (the decision makers for the child) to consent [13]. However, attempted eye salvage may negatively impact the family socioeconomically and psychologically, and still fail to save the eye.

Egypt is a low to middle income developing country [4,14,15] with low national income per capita, absence of a formalized health insurance system and limited national programs for financial support of families. Illiteracy is relatively high making cultural beliefs the dominant factor influencing decisions. The family is the main unit of the community and its continued union is important in any treatment plan. We now evaluate the financial, social and psychological impacts on families of treatment decisions for a child with retinoblastoma in the Egyptian setting.

Methods and materials

Study subjects

This research study was approved by the ethics committee of Alexandria University and is in accordance with the guidelines set forth by the Declaration of Helsinki.

A retrospective review was performed of the records of all patients who presented with unilateral retinoblastoma and were treated in Alexandria Main University Hospital from May 1st 2005 to May 1st 2013. Children with IIRC group C, D, or E eyes were included; children with clinical or radiological evidence of extraocular retinoblastoma at presentation, or IIRC group A or B eye were excluded. Data collected included presenting age, sex, family history, IIRC group, applied treatment modalities, side effects and complications of therapy and the final outcome regarding ocular and life salvage.

For any enucleated eye (whether primary or secondary), presence of high-risk histopathological features (defined as the presence of post-laminar optic nerve invasion and/or massive choroidal invasion, TNM stage pT3) and subsequent treatments were documented.

The duration of active treatment (time from diagnosis to 2 months after last treatment for active tumor) was measured for each child. For children still under active treatment, duration was measured up to May 1st 2013.

Two groups of children with advanced intraocular disease (IIRC groups C, D and E) were compared, depending on primary treatment:

- enucleation: treated by primary enucleation;
- salvage treatment: treated by any means to attempt ocular salvage.

The salvage treatment group was further subdivided according to the IIRC group of the eye. All IIRC group C (6) and 26 IIRC group D eyes were in the salvage treatment group.

Socioeconomic scoring

An ordinal scoring system was constructed to classify and compare the main socioeconomic and psychosocial impacts of treatment decisions between groups (i.e. this is not a measurement scale but data classification groups). Six parameters (financial burden, and financial, psychological, social, medical and tumor impact) were quantified by assigning a score for negative impact from 0 to 3 (Table 1). The total score ranged from 0–18. The score was considered 'high' if it was more than 50% (> 1.5/3 in each parameter and > 9/18 in the total score). The higher the score, the higher was the negative socioeconomic and psychosocial impact on the family.

In pilot studies as we developed the Socioeconomic score, it was suggested that enucleation would add to the social burden, for which we scored stigma and cosmetic outcome. We performed primary implant placement in all cases of primary enucleation with rapid, good cosmetic outcomes that did not lead to a burden of stigma in any child. Therefore, we did not include stigma and cosmetic outcome in the final score.

Statistical analysis

Data processing and analysis were performed using Statistical Package for Social Sciences (SPSS version 18). The level of significance was 5% (P < 0.05). For quantitative variables, the mean and standard deviation were calculated using the non-parametric Mann-Whitney test. For qualitative variables, Chi² test and Monte Carlo test were used. Spearman rank correlation test was used to assess the linear relationship between duration of follow-up of patients and the total Socioeconomic score. Simple linear regression analysis was performed to determine the effect of the duration of follow-up as an independent predictor for the total score.

Results

Case demographics

Sixty-six children with unilateral retinoblastoma were reviewed; 60 children met the inclusion criteria (unilateral intraocular retinoblastoma, IIRC groups C, D, E). Six children were excluded (2 extraocular disease; 4 IIRC group A or B, 3 with family history, median age at diagnosis 8 months).

Thirty-six (60%) children were female. The median age at diagnosis of the study subjects was 22 months (IQR 12–33). Two of the included children (3%) had positive family history, but were not seen until they had group C disease at 18 months of age and group D disease at 28 months of age, since their parents were unaware of risk for their children to develop retinoblastoma.
The eyes of the 60 children were classified as IIRC group C (6, 4%), D (27, 42%) or E (27, 42%) (Table 2). All group E eyes were primarily enucleated, with implant, without attempted salvage and was included in the enucleation study group; which included also one child with a group D eye that was primarily enucleated due to parental rejection of salvage treatment. The remaining 26 group D eyes and 6 group C eyes were included in the salvage treatment group. Absolute refusal of enucleation was not encountered in our series; however, an initial denial and search for a second opinion occurred in 7 cases (25%), but did not affect the timing of surgery for any child. All children showed acceptable cosmetic outcome with porous hydroxyapatite implant with either myo-conjunctival closure (17 children), or muscle suturing in front of the implant (11 children) with one delayed implant rejection requiring implant replacement, that did not affect the final cosmetic outcome.

**Ocular salvage**

In the salvage treatment group, primary therapy included systemic chemotherapy (vincristine, carboplatin and etoposide for 4–6 cycles) with focal therapy (31/32) or external beam radiation therapy (EBRT) (1/32). Further interventions for tumor control included second line chemotherapy (cyclophosphamide and doxorubicin for 4 cycles) (2), brachytherapy (5), EBRT (4), and intra-arterial chemotherapy using melphalan (9) (Table 2).

Despite attempted salvage, 78% (25/32) of eyes were eventually enucleated. The ocular salvage rate was 66% (4/6) for IIRC group C eyes and 11.1% (3/27) in IIRC group D eyes. Of the secondary enucleations, 44% (11/25) were because of failure to control tumor, and 56% (14/25) were because of financial, social or psychological impact of cumulative therapies, rather than failure of tumor control (Table 3).

### Active treatment duration

In the enucleation group, the median treatment duration was 2.2 months (IQR 1.8–2.2). In the salvage treatment group, overall treatment duration was median 14 months (IQR 8–20), significantly longer than the enucleation group (P < 0.01, Mann-Whitney test). For salvaged eyes (7/32),
Table 2  Case demographics and treatments.

<table>
<thead>
<tr>
<th></th>
<th>All</th>
<th>Enucleation</th>
<th>Salvage treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at diagnosis months, median (IQR)</td>
<td>26 (14–41)</td>
<td>21 (12–28)</td>
<td></td>
</tr>
<tr>
<td>Sex</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>24 40%</td>
<td>14 50%</td>
<td>10 31%</td>
</tr>
<tr>
<td>Female</td>
<td>36 60%</td>
<td>14 50%</td>
<td>22 69%</td>
</tr>
<tr>
<td>Total</td>
<td>60</td>
<td>28</td>
<td>32</td>
</tr>
<tr>
<td>Positive family history</td>
<td>2 3%</td>
<td>0 0%</td>
<td>2 6%</td>
</tr>
<tr>
<td>IIRC groups</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Group C</td>
<td>6 0%</td>
<td>0 0%</td>
<td>6 19%</td>
</tr>
<tr>
<td>Group D</td>
<td>27 1%</td>
<td>26 8%</td>
<td></td>
</tr>
<tr>
<td>Group E</td>
<td>27 96%</td>
<td>0 0%</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>60</td>
<td>28</td>
<td>32</td>
</tr>
<tr>
<td>Primary treatment</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Enucleation</td>
<td>28 100%</td>
<td>na</td>
<td></td>
</tr>
<tr>
<td>Chemo, focal therapy</td>
<td>31 na</td>
<td>31 97%</td>
<td></td>
</tr>
<tr>
<td>EBRT</td>
<td>1 na</td>
<td>1 3%</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>60</td>
<td>28</td>
<td>32</td>
</tr>
<tr>
<td>Further interventions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Post-enuc chemo</td>
<td>3</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Post-enuc radiation</td>
<td>4</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2nd line chemo</td>
<td>2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Brachytherapy</td>
<td>5</td>
<td></td>
<td></td>
</tr>
<tr>
<td>IAC</td>
<td>9</td>
<td></td>
<td></td>
</tr>
<tr>
<td>EBRT</td>
<td>4</td>
<td></td>
<td></td>
</tr>
<tr>
<td>BMT</td>
<td>2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Outcomes</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ocular salvage (2 active treatment)</td>
<td>7 12%</td>
<td>0 0%</td>
<td>7 22%</td>
</tr>
<tr>
<td>High-risk path</td>
<td>10 17%</td>
<td>4 14%</td>
<td>6 24%</td>
</tr>
<tr>
<td>Metastasis</td>
<td>6 12%</td>
<td>1 3.6%</td>
<td>5 16%</td>
</tr>
<tr>
<td>Alive</td>
<td>58 97%</td>
<td>27 96%</td>
<td>31 97%</td>
</tr>
</tbody>
</table>

IIRC: International Intraocular Retinoblastoma Classification; chemo: systemic chemotherapy; FT: focal treatment; EBRT: external beam radiation therapy; Brachy: brachytherapy; IAC: intra-arterial chemotherapy; na: not applicable; BMT: bone marrow transplantation; HRHPF: high-risk histopathological features; EO: extraocular. Bolds are totals and italics are percentages.

Table 3  Causes of secondary enucleation decision in the salvage treatment group.

<table>
<thead>
<tr>
<th>Cause of secondary enucleation</th>
<th>International intraocular retinoblastoma classification</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Group C (n=2)</td>
</tr>
<tr>
<td>Failure of tumor control despite prompt therapies</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Poor parental compliance for treatment and follow-up</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Financial problems prevented proper therapy implementation</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Social problems affecting family unity with struggles on treatment</td>
<td>1 (50%)</td>
</tr>
<tr>
<td>Mental exhaustion of parents from continuing hope of ocular salvage</td>
<td>1 (50%)</td>
</tr>
</tbody>
</table>
median treatment duration was 19 months (IQR 16–22), longer than median treatment duration of 13 months (IQR 7–19) for non-salvaged eyes, but that difference was insignificant (P = 0.184, Z = −1.328, Mann-Whitney test). Two IIRC group C eyes were still under active treatment for peripheral tumor recurrences.

**Histopathology outcome**

In the enucleation group, 4/28 (14.3%) eyes showed high-risk histopathological features [11,12]. In the salvage treatment group, 6/25 (24%) eyes eventually enucleated showed high-risk histopathological features (all IIRC D), with one (4%) showing definite extraocular spread. Identification of high-risk pathological features was followed by adjuvant systemic chemotherapy with or without EBRT.

**Extraocular or metastatic morbidity and mortality**

In the enucleation group, one child of 28 (3.6%) who had received post-enucleation chemotherapy because of high-risk histopathological features, developed orbital and central nervous system (CNS) metastasis that was treated with additional radiation therapy, but the child died.

In the salvage treatment group, 5/32 children (15.6%) developed extraocular or metastatic disease [cerebrospinal fluid (CSF) positive tumor cells in 3/5 children]. Three children had high-risk histopathological features in the secondarily enucleated eye, and received chemotherapy. One late enucleated eye showed no high-risk histopathological features so did not receive additional treatment, but later showed CNS metastasis with CSF positive for tumor cells, without a definite CNS mass. The fifth child was on active treatment for ocular salvage but was temporarily lost to follow-up, during which time she received two intravitreal bevacizumab injections elsewhere, then she returned with extraocular disease and died 7 months later.

All five children with extraocular disease received adjuvant chemo-radiotherapy and one child underwent bone marrow transplantation. Three children are still under active treatment for metastatic or extraocular disease (Table 2). Metastasis was more frequent in the salvage treatment group (5/32) than in the enucleation group (1/28), this was not significant (Fisher’s Exact test, P = 0.201).

The 5 children in the salvage treatment group who developed metastasis had been under active treatment for a median duration of 19 months (IQR 19–23) while 27/32 (84%) with no extraocular disease were under active treatment for a median of 12 months (IQR 7–19). This difference was statistically significant (P = 0.02, Z = −3.048, Mann-Whitney test).

**Socioeconomic Impact score**

The salvage treatment group had a median Socioeconomic score of 11 (IQR 9–13), significantly higher (higher negative impact) than median 4 (IQR 2–6.5) for the enucleation group (P < 0.01, Mann-Whitney test) (Table 4). Scores for financial burden, financial impact, psychological and social impacts were high (> 50%) in the salvage treatment group, while the enucleation group received no high scores for any parameter. All parameters except for the psychological impact were significantly higher (P < 0.01) in the salvage group (Table 4). Psychological impact was not significantly different (P = 0.62) with a trend to being lower in the enucleation group.

In the salvage treatment group, both IIRC groups C and D eyes showed high financial impact and burden scores (4.5/6 and 5/6, respectively). All the remaining parameters were significantly (P < 0.01, Monte Carlo test) lower in IIRC group C than group D eyes (Table 4).

The Socioeconomic scores were significantly higher for the 5 families whose children developed metastatic disease (median score 17, IQR 14–17) with the longest duration of treatment (median 19 months, IQR 19–23), compared to the 27 families whose children had no metastatic disease (median 10 months, IQR 9–13) and treatment duration (median 11 months, IQR 7–16) (P < 0.01, Mann-Whitney test). Also, the score was significantly higher in cases of metastasis regardless of the treatment group (P < 0.01, Mann-Whitney test).

There was a positive correlation between the duration of follow-up (months) and the total Socioeconomic Impact score regardless of the study group (r = 0.69, P < 0.01). Simple linear regression analysis revealed that each additional month of active treatment increased the total score by 0.371 points (P < 0.01). The Socioeconomic Impact score showed no statistical difference with presenting age or sex.

**Discussion**

The diagnosis of a child with retinoblastoma carries significant economic, psychological and social impacts on families [16]. This is intensified with the dilemma of choosing between saving or removing an affected eye.

For bilateral retinoblastoma, trial ocular salvage for IIRC group D eyes is standard, in hope of saving at least one eye. However, for advanced intraocular unilateral eyes (groups D and E eyes) trial ocular salvage may not be justified as the other eye is perfectly normal and enucleation is curative in 93% of unilateral cases [3,8].

Enucleation has been for years the mainstay of therapy for advanced intraocular unilateral retinoblastoma, and EBRT was rarely used due to risk of secondary cancers and poor cosmetic outcome [3,8,17]. Introduction of systemic chemotherapy [18] selective intra-arterial chemotherapy [9,10,19] focal chemotherapy and stereotactic radiation [20,21] has resulted in widespread advocacy for attempted ocular salvage, without evaluation of impact on the child and family affected by unilateral retinoblastoma.

Eye salvage attempt for retinoblastoma is expensive [22] with long treatment courses that require frequent and careful follow-up, mostly under anesthesia. In other serious conditions, financial burden has been shown to affect decisions and outcomes [23–25] for retinoblastoma, financial burden has not been deeply studied, and may be missing from informed discussions with parents. In many developed countries, the affected children have health insurance, so costs are a lesser burden to families. In countries such as Egypt, with low national income per capita income and limited health insurance coverage, we show that the financial burden is important in the treatment decisions.
Table 4 Socioeconomic scores of the enucleation and salvage treatment groups.

<table>
<thead>
<tr>
<th>Parameter (median scores)</th>
<th>Enucleation Group C (n = 6)</th>
<th>Group D (n = 26)</th>
<th>Total (n = 32)</th>
<th>Test of significance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Financial burden</td>
<td>0.5</td>
<td>2.5</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Financial impact</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>Psychological impact</td>
<td>1</td>
<td>1</td>
<td>2</td>
<td>1.5</td>
</tr>
<tr>
<td>Social impact</td>
<td>1</td>
<td>1</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Medical impact</td>
<td>0.5</td>
<td>0.5</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tumor impact</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0.5</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total (0–18)</td>
<td>4</td>
<td>7</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td></td>
<td></td>
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</tbody>
</table>

IIRC: International Intraocular classification. Bold are the totals of socioeconomic score and italics are always the P-value.

* Statistically significant.

There are many other factors to be taken into account in full consideration of the choice between attempted eye salvage and primary enucleation in order to fully inform the discussion with parents about primary treatment choices. Parental compliance throughout the treatment course is essential, or life of the child can be at risk. Also, repeated anesthetics are now recognized to incur neurodevelopmental risks on young children [26–28].

Delayed enucleation exposes the child to risk of metastasis and masks appreciation of high-risk histopathology, especially for group E eyes, which display clinical features suggesting real or imminent extraocular disease [24]. This very serious consequence of attempted eye salvage in advanced eyes is reiterated by our study: metastasis occurred in 1/28 (3.6%) children who had primary enucleation, but 5/32 (16%) who had attempted eye salvage, associated with long treatment durations and higher Socioeconomic Impact scores.

Social stigma of loss of an eye is often cited as a reason to try to avoid enucleation together with fear of visual disability [13]. Parental refusal of enucleation varies widely, ranging from 2% to 100% [13,17,29]. The loss of a child’s eye can have psychological implications for their parents [16]. It has been suggested that the stigma of enucleation is an indirect consequence of the parent’s inability to cope with a perceived ‘damaged’ child, rather than a direct effect of the surgery on the child themselves [16]. More psychological support, rather than turning to conservative therapies, may be beneficial.

Indeed, quality of life of retinoblastoma survivors is comparable to unaffected individuals of the same age [30]. Survivors of enucleated unilateral retinoblastoma who undergo primary enucleation reportedly experience better psychological outcomes and quality of life than bilateral retinoblastoma survivors, who presumably undergo complex conservative therapy [31]. We did not encounter absolute enucleation refusal; however initial denial and search for a second opinion occurred in a few instances, without significant delay in eventual enucleation. It has been shown that increased time spent with parents to comprehensively educate them about enucleation and its consequences may help avoid enucleation refusal [29].

Our study found that enucleation had a low negative socioeconomic impact, supporting the value of enucleation over salvage treatments for unilateral advanced intraocular retinoblastoma [31,32]. Wilson et al. [33] studied the impact of attempted salvage treatments in a developed country, and concluded ‘Successful retinoblastoma management requires close surveillance, aggressive consolidation, and numerous anesthetic procedures, all of which the patients and the families must endure’. The psychological and social impact on the families was not measured.

The diagnosis of retinoblastoma is stressful for the parents who seek responsible treatment decisions of behalf of their children [33,34]. Hope to save an eye, however probable or unrealistic, motivates the parents and may help them to cope in the short term. However, fully informed consent to treatment would include best estimate of various risks and benefits. For the choice between primary enucleation and attempted eye salvage, informed consent would ideally disclose all variables with best estimate of likelihood for success to keep the eye and other impacts on the child and
family. In our study, 56% of the eyes eventually enucleated despite attempted eye salvage, were enucleated because of negative socioeconomic impacts, rather than failure of therapy. This contributed to our low ocular salvage of 22%. Our study documents the socioeconomic impacts of treatment decisions in unilateral advanced intraocular retinoblastoma using a retrospective score on a relatively large number of families. To expand this data, we propose a prospective study of fully informed discussion of the choice between primary enucleation or attempted eye salvage including potential socioeconomic impact, with measurement of outcomes. We recognize that since our study was applied in a single community, the results may be limited by financial, social and cultural differences of our community, and not widely generalizable. Finally, we did not look at how demographic factors might have affected scores, such as parental level of education, baseline socioeconomic level, and broader network of family support, family size, and duration of treatment and follow-up.

Conclusions

To our knowledge, this is the first study of socioeconomic impacts of retinoblastoma therapy decisions. We found that primary enucleation for unilateral advanced intraocular retinoblastoma was associated with fewer negative financial, social and psychological impacts on families than attempted ocular salvage. In developing countries with similar conditions as Egypt, primary enucleation serves the child and family well.

Disclosure of interest

The authors declare that they have no conflict of interest concerning this article.

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