Flash-lamp pulsed-dye laser treatment of port-wine stains in childhood. A case of technology assessment

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Assessment of the outcome of laser treatment of children with a facial port-wine stain

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Submitted for publication
Assessment of the outcome of laser treatment

Abstract

Background - The flash-lamp pulsed-dye laser (FPDL) is now an established form of treatment of port-wine stains (PWS) in childhood. Few studies so far regarded patients' assessments of laser surgery. None of them studied the assessment of the outcome of therapy during childhood.

Methods - We prospectively studied, 52 consecutive parents of patients (aged 0 to 13) with a facial PWS. Parents were asked to evaluate their expectations of therapy and outcome of therapy of their children. During FPDL treatment an evaluative questionnaire, completed by the children's parents, was distributed.

Results - Seventy-seven percent of the parents of children responded. Children went through a mean treatment period of 2.4 years. On average children aged 0 to 5 and children aged 6 to 13 year had gained 30% and 43% clearance respectively. At the beginning of treatment 60% of the parents reported that they would accept a negative treatment outcome. The majority of the parents reported that the received information prior to treatment was adequate. Fifty percent of the parents would have preferred to receive more information about the duration of treatment. Most parents (90%) felt that the PWS of their child had become less striking as a result of therapy. Two thirds reported that they were overall satisfied with the result of FPDL treatment of their child. Parents of younger children were willing to pay more for the laser treatment of their child compared to parents of teenagers.

Conclusions - Although clearing is partial and treatment of children takes considerably more time than anticipated, parents of patients are satisfied with the results of therapy.
**Introduction**

The flash-lamp pulsed-dye laser treatment (FPDL) is now an established form of treatment for facial port-wine stains (PWS). It is the preferred option in children and some have argued that treatment should start as early as possible.¹⁻⁶

So far the outcome of FPDL treatment and the eventual adverse effects have been evaluated by eliciting overall statements of success or failure from unblinded clinicians. Such a global assessment is vulnerable for bias and cannot escape a certain degree of subjectivity, as it has to be based on an impression of the disfiguring nature of the PWS and its modifications under laser treatment.⁷⁻⁸ Such an appraisal requires a perceptual judgement of the changes in color and size of the PWS and the differences in surface texture.⁹⁻¹²

In the management of PWS it is not the stain as such that warrants treatment, but the consequences of the disfiguring aspects of the PWS on the psychosocial well-being of the bearer.

The treating physicians may not always be the most appropriate observers to evaluate these consequences, compared to the patients themselves or their parents. As expert witnesses of care, patients and parents hold a unique vantage point that can be of help to decision makers in health care. Based on these arguments, it can be argued that any assessment of FPDL treatment, both of the procedure itself and of its outcome, should therefore at least be accompanied by the judgement and opinion of patients and their families.

There are other reasons to call for a more systematic consultation of participants' opinion in laser treatment. Patients and their parents can have unrealistic expectations of the eventual outcome of therapy, hoping for a complete disappearance of the stain where such an outcome is out of reach.¹³ In order to prevent disappointment and dissatisfaction, measuring patient-based outcome, including the acceptance and satisfaction with the received information and the results of treatment, can help clinicians in evaluating their (individual) patient care and informing future patients.

Few authors so far have looked at patients' assessment of laser surgery. Their studies reflect the assessment of adult patients¹⁴ and evaluated the results of other laser types¹⁵⁻¹⁷, different anatomical sites¹⁷,¹⁸, mostly related to a short treatment period.
Assessment of the outcome of laser treatment

In this prospective study we report the outcome of laser treatment of facial PWS in young children from a parents’ point of view. A parent-report questionnaire was developed, covering the following topics: treatment expectations, adequacy of the received information on treatment, global satisfaction with present appearance, change in facial appearance, and an appraisal of the achieved result in economic terms. Also included were two measures of behavioral intentions. Parents’ responses were seen as an expression of parental perceptions of their child’s adjustment to the results of treatment, which may depend on the age of the child. For this reason, results were compared between age groups. In addition, we analyzed to what degree the variability in satisfaction and willingness-to-pay could be explained by differences in treatment characteristics and outcome.

Material and methods

Patients

Fifty-two consecutive patients (aged 0-13) with previously untreated facial PWS were invited to this prospective comparative study. All patients were treated between December 1991 and March 1995 in the department of Reconstructive and Plastic Surgery in the Academic Medical Center at the University of Amsterdam, the Netherlands. Prior to treatment oral informed consent was obtained from the parents or caretakers. During treatment patients received a standard information briefing.

Treatment

Patients were treated with a Candela SPTL-1 flash-lamp pumped pulsed-dye laser. In most patients a single treatment of the entire PWS required a number of visits. The average period between 2 visits was 8.6 weeks (sd. 3.0).

During treatment a eutectic mixture of prilocaine and lidocaine (EMLA®-cream) and/or nerve blocks were used. General anesthesia in a day-care unit was used if children became highly anxious or pain sensitive.

The difference in color between the healthy skin and the PWS was measured with a colorimeter10 (a Minolta chromameter type CR-300). Color measurements were taken before laser treatment started and after each following treatment of the entire PWS. Color differences were estimated based on the color measurement of the PWS and a symmetrical area in the face.
Questionnaire

At the beginning of laser therapy the parents of the patients were asked to indicate their anticipated acceptance of a negative treatment outcome, i.e. no clearance of the PWS. Answers could be given on a five point Likert rating scale, ranging from "strongly agree" to "strongly disagree".

Between May-July 1995 we sent out paper questionnaires to the parents, with one reminder after 6 weeks. In this questionnaire, parents were first asked to answer five questions referring to the level of satisfaction with the amount of information received, as well as with the quality of the instruction about the clinical routine. The latter included discoloring of the PWS, pain, results, duration and the actual treatment procedure. Answers could be given on a five point Likert rating scale, ranging from "strongly agree" to "strongly disagree".

In addition parents were asked whether they felt satisfied with the results achieved so far with laser treatment of their child's PWS. They were invited to indicate perceived changes in the appearance of their child by marking one from three ordered categories.

Parents were subsequently asked to think back to the situation in which they had to make the choice to apply their child for laser therapy. They were requested to indicate how much they would be willing to pay for a single laser treatment of their child's PWS on a visual analogue scale. The scale was anchored on hypothetical payment prices. The anchors ranged from 0 to 1500 Dutch Guilders (DGL) (0 to app. 750 USD). No additional information about the risks and benefits of laser treatment of PWS in children was provided, so patients had to rely on their own experience with treatment of their child in answering this question. We also asked parents to indicate their net take-home salary.

We included two measures of behavioral intentions in the questionnaire. Patients were asked whether they would recommend laser treatment to (1) a personal friend with a child with a PWS and (2) to a personal friend with a PWS.
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Analysis

Patients were divided in two age groups: young children, ranging in age from 0 to 5 year, and children aged 6 to 13 year. We used the F statistic to compare the reduction in color differences between the two age groups.

Responses to the five-point scales were re-grouped into positive (4 or 5 on the scale), intermediate (3 on the scale) and negative (1 or 2 on the scale) answers. Close-ended responses were summarized using descriptive statistics.

The significance of differences in willingness-to-pay between the two age groups was assessed using the Mann-Whitney U test statistic. The influence of patient-parent characteristics and actual treatment outcome on the reported global satisfaction and willingness-to-pay amounts was examined using correlational analysis. All calculated P values were two-tailed, with values of .05 or lower indicating statistical significance.

Results

Patients

The parents or caretakers of 40 children (77%) returned the questionnaire: 21/25 patients aged 0-5 and of 19/27 patients aged 6-13. Thirty of the questionnaires were completed by the mothers (75%), 7 by the fathers (18%) and 3 were completed by both parents.

In the younger group (0-5 year) the parents of 6 boys and 15 girls responded. At the start of the treatment the mean age of the children included in this group was 3.5 years (sd. 1.5). The average facial surface area of their PWS was 71.8 cm² (sd. 60.9).

In the older group (6-13 year) 9 parents of male patients and 10 parents of female patients returned the questionnaire. The mean age before treatment of the children included in this group was 7.5 (sd. 1.5). The average surface area of their PWS was 117.6 cm² (sd. 148.0).

Treatment

Table 1 contains the general treatment characteristics of the respondents. The mean treatment period at the time of this study was 2.4 years. In this period patients had received an average of 8 total treatments of the PWS. Overall a
Table 1. Treatment characteristics.

<table>
<thead>
<tr>
<th></th>
<th>0-5 year (n=21)</th>
<th>6-13 year (n=19)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Average visits per patient(^1)</td>
<td>15 (6)</td>
<td>14 (7)</td>
</tr>
<tr>
<td>Total treatments of the entire PWS [range]</td>
<td>8.2 (2.3) [5 – 13]</td>
<td>7.9 (3.0) [1 – 13]</td>
</tr>
<tr>
<td>Treatment period (year)</td>
<td>2.7 (0.5)</td>
<td>2.1 (0.7)</td>
</tr>
<tr>
<td>Number of pulses per visit (^1)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>With general anesthesia (n=14)</td>
<td>182 (267)</td>
<td>201 (268)</td>
</tr>
<tr>
<td>Without general anesthesia (n=26)</td>
<td>571 (433)</td>
<td>819 (766)</td>
</tr>
</tbody>
</table>

\(^1\) mean (standard deviation)

A smaller average number of laser pulses per visit had been administered in the younger age group (0-5 year).

General anesthesia in a day-care unit had been introduced during treatment for 14 (35%) children: 11/21 in the younger age group (0-5 year) and 3/19 in the older age group (6-13 year). Before the introduction of general anesthesia a mixture of prilocaine and lidocaine (EMLA\textsuperscript{®}-cream) in combination with midazolam (Dormicum\textsuperscript{®}) had been used in 9 of these patients. All other children applied EMLA\textsuperscript{®} before treatment. We learned from the clinicians that distress and the low pain threshold of these children were factors influencing the decision to use general anesthesia. Although both factors were related to the size and the location of the PWS, there was no general policy favoring local anesthetics in small lesions and general anesthesia in large lesions.

The color differences at the initiation of treatment between the PWS and the contra-lateral healthy skin were comparable between both age groups (F=2.0, p=0.16). On average children aged 0 to 5 year and those aged 6 to 13 year achieved a relative improvement of 30% (sd. 33) and 43% (sd. 22) respectively. This difference was not statistically significant (p=0.13).
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**Table 2. Global assessment of clinical outcome.**

<table>
<thead>
<tr>
<th>Parents of patients</th>
<th>0-5 year</th>
<th>6-13 year</th>
</tr>
</thead>
<tbody>
<tr>
<td>(n=21)</td>
<td>(n=19)</td>
<td></td>
</tr>
<tr>
<td>I am satisfied with the results of laser treatment for my child’s PWS</td>
<td>15</td>
<td>13</td>
</tr>
<tr>
<td>The PWS has disappeared</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>The PWS is less striking</td>
<td>20</td>
<td>17</td>
</tr>
<tr>
<td>The PWS has not changed</td>
<td>-</td>
<td>1</td>
</tr>
</tbody>
</table>

**Questionnaire**

At the beginning of treatment 24/40 of the responding parents (60%) announced that they would accept a negative outcome, i.e. no clearance of the PWS.

Most parents reported that they had received adequate information about discoloring of the PWS after treatment (36/40), pain associated with treatment (36/40), the treatment procedure itself (39/40), and the expected results of laser treatment (35/40). Fifty percent of the respondents (20/40) indicated that the information about the duration of the treatment was adequate; whereas the other half would have preferred receiving more specific information about this issue.

The answers to the global assessment items in the follow-up questionnaire that refer to the parent/patient-based outcome of laser therapy are summarized in Table 2. Two thirds of the parents (28/40) indicated that they were overall satisfied with the result of the laser treatment of their child. Of the respondents group 30% (12/40) said they were not satisfied with the result.

Two parents felt that the PWS of their child had completely disappeared as a result of laser treatment. Their children, aged 0-5 and 6-13, had at that point received 10 and 4 total treatments of the entire PWS, respectively. In the older age group the parents of one child felt that the PWS of their child had not changed after 6 total treatments of the entire PWS. The majority of the parents (37/40, 90%) felt that the PWS of their child had become less striking as a result of laser treatment. We did not find an association between surface area of the PWS, age of the child, clearing of the PWS and overall satisfaction.
with the result of treatment (Table 3). In contrast, parents of patients who had received a high number of total treatments of the entire PWS expressed less satisfaction with outcome.

Parents were also asked to assign the economic value of laser treatment and the achieved treatment results, based on their current experience. Four parents, two in the younger and two in the older age group did not want to answer this virtual question on willingness-to-pay (WTP). They saw it as the responsibility of society to pay for the treatment. The median amount parents of children in the younger age group were willing to pay was DGL 300,- (interquartile range: DGL 135,- — 1500,-; app. 78 USD — 750 USD). In the older age group, the corresponding amount was DGL 250,- (interquartile range: DGL 60,- — 950,-; app. 30 USD — 475 USD) (Figure 1). This difference in willingness-to-pay was not statistically significant (p=0.19). Five parents of children (24%), aged 0-5 year, and three parents of children (16%) aged 6-13 year were willing to pay the highest economic price mentioned on the scale (DGL 1500,-; app. 750 USD).

Figure 1. Median amounts (DGL) of Willingness-to-Pay with interquartile ranges.
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Table 3. Correlation between Parent-patient characteristics, Treatment outcome, Satisfaction and Willingness-To-Pay.

<table>
<thead>
<tr>
<th>Predictors</th>
<th>Satisfaction (n=40)</th>
<th>Willingness-To-Pay (n=36)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>R</td>
<td>p-value</td>
</tr>
<tr>
<td>Parent-patient characteristics</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Surface area of the PWS</td>
<td>-0.07</td>
<td>0.68</td>
</tr>
<tr>
<td>Age of the child</td>
<td>-0.08</td>
<td>0.64</td>
</tr>
<tr>
<td>Clearing of the PWS</td>
<td>0.03</td>
<td>0.85</td>
</tr>
<tr>
<td>Total treatments</td>
<td>-0.36</td>
<td>0.03</td>
</tr>
<tr>
<td>Treatment outcome</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Clearing of the PWS</td>
<td>0.07</td>
<td>0.67</td>
</tr>
<tr>
<td>Total treatments</td>
<td>0.23</td>
<td>0.19</td>
</tr>
</tbody>
</table>

The average reported family income did not differ between age groups. Families with higher incomes were willing to pay more for treatment. We did not find an association between surface area of the PWS, age of the child, PWS clearing, total treatments of the entire PWS and the reported WTP amount (Table 3).

All 21 parents of the younger children and 17 out of 19 parents of the older children would recommend laser treatment to a friend or to the child of a friend.

Discussion

This study shows that it is feasible and informative to ask parents for an evaluation of treatment and its outcome through a structured questionnaire. Results demonstrate a considerable level of parent satisfaction after laser
treatment of a facial PWS in their children, even though optimal clearing had not been obtained.

Our results also show room for improvement, for example in the quality of information about expected treatment duration. Some differences were observed between age groups, although these were not statistically significant. When asked how much money they were willing to pay for one laser treatment of their child, parents in the youngest age group (0-5 year) reported higher amounts than parents in the older age group (6-13 year).

The results shown cannot unconditionally be generalized to other conditions and settings. Our study, based on a small sample, only included patients and parents who had chosen to pursue laser therapy. Parents that had made an informed decision not to opt for laser treatment could not be included. Sixty-three percent of our patients were female and 37% male, while there is supposed to be an equal sex distribution among patients with PWS. It is possible that parents of female patients value facial appearance higher and consequently seek treatment more actively. We should also keep in mind that it is unknown to what degree these assessments in parents of young children are stable over time.

We found a negative association between level of global satisfaction with treatment outcome a number of total treatments of the entire PWS received. None of the patients in this study had completed treatment at the point where outcome of laser therapy was evaluated. It is possible that feelings of satisfaction become stronger with the gradual disappearance of the PWS. On the other hand, as maximal clearing takes multiple treatments, dissatisfaction may grow with the increasing burden of a prolonged treatment duration.

In this study, we found similar outcome across age groups. It has been suggested that the periods of family stress accumulate as the child with PWS grows, with the major stress for the parents occurring during the early years of the child. Our results point in this direction, although none of the differences, considered in isolation, were statistically significant. Parents of younger children were willing to pay more for treatment compared to parents of teenagers. The higher anticipated benefit of PWS clearance at an early age could be held responsible for this difference.

The assessment of the FPDL treatment of children as reported by parents is likely to reflect the parents own opinion and feelings. People see illnesses as subtracting value from their lives, and parents tend to be more protective of
their children than of themselves. Yet little is known about the parents of children with a PWS and the characteristics of their parent-child relationship. Variables that might influence the decision of parents to seek treatment for their child have not yet been identified. In a recent study of 70 adult patients applying for FPDL treatment, participants reported retrospectively that parents had been very concerned about their stain and that their parents had made many attempts at therapy of the PWS during their childhood.²⁴

Patient and parent satisfaction is a multidimensional concept. Satisfaction with treatment is not only determined by outcome. Whether patients and parents are satisfied will to a large extent be determined by the interplay between prior expectations and actual treatment outcome. The former can be influenced by the information provided by the responsible clinicians before treatment starts.²⁵ Willingness-to-pay for treatment is known to depend on similar factors, as well as on family income.²⁶ Kalick concluded that patients with a PWS had unrealistic perfectionist expectations of their argon laser treatment.¹³ Augustin et al., studying a group of 70 adult patients receiving FPDL, reported that about 90% had high or very high expectations that the stain would disappear with treatment.²⁴

Beyond prior expectations and outcome, many other features of the process of delivering health care can influence treatment satisfaction. Recently Miller et al. reported that parents of children with a facial PWS experienced less parental stress if they were satisfied with staff communication during treatment.²³ In our study the majority of the parents reported that they would accept a negative outcome of treatment and that they had received adequate information, although the information about expected treatment duration left room for improvement. We think that providing comprehensive and detailed information prior to the initiation of laser therapy can help in avoiding unrealistic expectations. Even when clearing is not optimal or when treatment takes considerably more time than anticipated, parents and patients may still be satisfied, if they feel that their own viewpoint has been taken into account and that an acceptable outcome has been reached.
References

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