

Original Investigation

Social Impact of Facial Infantile Hemangiomas in Preteen Children

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IMPORTANCE Involved infantile facial hemangiomas (IHs) may adversely affect the social skills of children.

OBJECTIVE To assess the social impact of involuted facial IHs, with or without prior treatment, in preteen children.

DESIGN, SETTING, AND PARTICIPANTS An observational, cross-sectional study of social anxiety and skills in preteen children with facial IHs diagnosed during infancy. The study took place in an academic institution and a community dermatology practice between January 1, 2013, and July 30, 2014. Records on 236 children with IHs located in a cosmetically sensitive area were identified; of those, 144 potential participants (parents) were reached by telephone and mailed study packets. Thirty completed questionnaires were returned. Data analysis was performed from August 1, 2014, to September 7, 2015.

INTERVENTIONS The questionnaires included the following psychiatric scales: (1) Social Anxiety Scale for Children-Revised (SASC-R), completed by parents and children, including the domains of Fear of Negative Evaluation and Social Avoidance/Distress in New Situations (SAD-New) (higher scores indicate greater social anxiety), and (2) Social Competency Inventory (SCI), completed by parents, including the domains of Prosocial Behavior and Social Initiative (lower scores indicate poorer social competency).

MAIN OUTCOMES AND MEASURES Demographics, clinical details, and survey responses were collected. Analysis was conducted using *t* tests to compare scores for each survey domain with established normative data and between sex as well as between treatment vs nontreatment groups.

RESULTS Of the 144 potential participants, 30 (21%) responded. The mean age of the preteen subjects was 10.0 years (range, 5.4-12.9 years) with a 2:1 female to male ratio. Twenty-five children (83%) had a single IH, and the remaining 5 participants (17%) had multiple IHs, with at least 1 IH in a cosmetically sensitive area. The periocular region was the most common site of the IH (10 [33%]), followed by the nose (6 [20%]), cheek (5 [17%]), forehead (4 [13%]), lip or perioral region (4 [13%]), and ear (1 [3%]). Eighteen children (60%) had received treatment for their IH. With results reported as mean (SD), the SASC-R test showed that social anxiety of the children was not increased over normative data; however, those who did not receive IH treatment had significantly greater anxiety for new situations compared with those who received treatment (SAD-New: 15.5 [5.1] vs 11.5 [3.8]; $P = .02$). Results of the SCI scale indicated that the Prosocial Orientation domain score for the children was similar to normative data (3.96 [0.48] vs 3.89 [0.55], $P = .50$). Social Initiative domain scores were significantly poorer in children who did not receive treatment vs those who received treatment (3.45 [0.43] vs 4.03 [0.55]; $P = .006$).

CONCLUSIONS AND RELEVANCE Preteen children with involuted, untreated facial IHs have higher Social Anxiety domain scores in new situations and decreased Social Initiative domain scores compared with children who receive treatment for facial IH. Although this study is limited by a small sample size, it raises important considerations for whether early treatment of facial IHs in cosmetically sensitive areas has a beneficial effect on social skills in preteens.

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Infantile hemangiomas (IHs) demonstrate a characteristic growth pattern of proliferation early in infancy followed by plateaued growth and ultimate spontaneous involution. Despite involution, complete regression is rare and residual scarring is characteristic, particularly for superficial skin lesions.¹ Most IHs occur on the head and neck, usually in conspicuous and cosmetically sensitive regions. As a result, it is important to consider the effect that facial scars of IHs may have on children during critical periods of social development.

Before the serendipitous discovery of propranolol hydrochloride as an effective treatment, corticosteroids were the mainstay of IH therapy. Although reversible, the high rate of adverse events limited corticosteroid use to treatment of symptomatic lesions. The higher safety profile of propranolol is apparent² and is expanding our indications to treat asymptomatic lesions in cosmetically sensitive areas in an effort to improve the ultimate outcome of involution.

Previous research investigating the psychosocial effect of IHs found that parents of affected children reported on negative comments, such as stares from strangers and accusations of child abuse.³ In addition, parents expressed a belief that their child's life would have been different without an IH and demonstrated concern for their child's social functioning. Another study using Joseph's preschool and primary self-concept test⁴ demonstrated that children aged 3 to 5 years with IHs of the face, head, or neck felt more negatively valued than their peers without IHs. Clearly, the altered physical appearance due to IHs may produce behavioral and self-conceptual responses in children. Despite these studies, the psychosocial effect of facial IHs in young children remains controversial. A quality-of-life study⁵ conducted in children aged 5 to 8 years in an Israeli medical center concluded that head and neck IHs do not have a negative effect on quality of life or self-esteem; however, 43% of the lesions were on the scalp or the neck, where IHs may not cause the same cosmetic concerns as those on the face, such as those in the present study. Furthermore, patients with scars had significantly lower self-esteem scores, suggesting that cosmetic deformity may have a negative effect on self-esteem.

In this observational cohort study, we explored the social impact of facial IHs in cosmetically sensitive areas in preadolescent children aged 8 to 13 years using the following validated psychiatric instruments: (1) Social Anxiety Scale for Children-Revised⁶ completed by parents (SASC-R/Parent) and by children (SASC-R/Child), with the latter optional for study enrollment, and (2) Social Competence Inventory⁷ (SCI) completed by the parents.

Methods

Study Design and Participants

After obtaining institutional review board approval from the Children's National Medical Center the records of all 665 children born between January 1, 2000, and December 31, 2005, with a diagnosis of a hemangioma (*International Classification of Diseases, Ninth Revision [ICD-9]* code 228) at Children's National Medical Center or Virginia Dermatology Practice were re-

viewed to identify children who had lesions in these cosmetically sensitive regions: periocular, cheek, nose, ear, lip, and forehead. Those with scalp and neck lesions were not included because such lesions are easily camouflaged. Study packets were mailed to all eligible participants after they were contacted by telephone to explain the study and verify the mailing address. Updated contact information was also sought via searches using Google, White Pages, and Spokeo. Study packets included the following documents approved by the institutional review board: a letter describing the study, consent and assent forms, 3 questionnaires (SCI, SASC-R/Parent and SASC-R/Child), and a demographic/health information form. Parents were informed that their child's completion of the SASR-C/Child was optional if they thought the questions would disturb the child. Eligible participants who did not return surveys within 3 months were recontacted by telephone and study packets were re-sent. Although not required for study participation, parents were asked to send a recent photograph of their child that showed the involuted IH. Participants returning the requested information were sent gift cards.

SASC-R Questionnaire

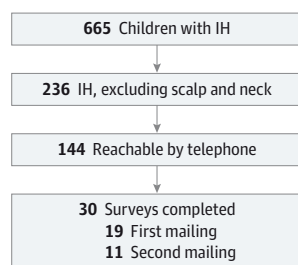
The SASC-R assesses social anxiety in preadolescent children.⁶ Both the SASC-R/Parent and SASC-R/Child surveys consist of the same 18 Likert-scale items plus 4 filler items worded appropriately for the responder's age. The Likert scale ranges from 1 (not at all) to 5 (all the time), with higher scores indicating greater social anxiety. Children with social anxiety have significantly higher scores compared with nonsocially anxious children.⁸ The total score (sum) is calculated for each domain:

1. Fear of Negative Evaluation (FNE) includes 8 questions (maximum sum score, 40) representing the child's subjective experience of anxiety. Questions include, "I worry about being teased" and "I'm afraid that others will not like me."
2. Social Avoidance and Distress-General (SAD-General) includes 4 questions (maximum sum score, 20) examining behavioral consequences of anxiety experienced more generally in the presence of peers. Questions include, "It's hard for me to ask other kids to do things with me" and "I feel shy even with kids I know well."
3. Social Avoidance and Distress-New (SAD-New) includes 6 questions (maximum sum score, 30) assessing behavioral consequences of anxiety experienced in light of new situations or unfamiliar peers. Questions include, "I only talk to kids I know really well" and "I get nervous when I meet new kids."

Social Competence Inventory

The SCI comprises 25 items designed to measure parents' perception of their child's ability to engage in social settings.⁷ A child's version of the survey does not exist. The survey responses are based on a 5-point Likert scale (1 indicates never; 5, always), with 6 of the responses requiring reverse scoring before assessment. Unlike the SASC-R, the SCI survey assesses the mean score, and a lower score is associated with poorer social competency. The mean scores are calculated for the following 2 domains:

Figure 1. Enrollment



Study enrollment of 8- to 12-year-old children with a history of facial infantile hemangioma (IH).

1. Prosocial orientation (17 questions, minimum mean score, 1) assessing a child's voluntary social behavior, such as helping or sharing, intended to benefit another. Questions include, "My child tries to comfort a peer who is upset, not feeling well, or has been hurt" and "My child is able to bx; interpret (decode) another child's feelings if he/she is happy, angry, or sad."
2. Social initiative (8 questions, minimum mean score, 1) assessing a child's willingness to engage in social interactions. Questions include, "My child easily makes contact with unfamiliar children" and the reverse-scored question, "My child is withdrawn from peers."

Statistical Analysis

Scale reliability was assessed using Cronbach α with a cut-off value of >0.70 . Participants' responses were compared with established normative data (historic controls) using a paired t test, with $P < .05$ considered statistically significant. The t test was also used to determine differences based on sex and history of prior IH treatment.

The photographs of the children were assessed by 3 surgeons with vascular anomaly expertise who rated the cosmetic effect of the IH as major if they would offer surgical intervention for the child if requested or minor if they would not offer surgical intervention if requested.

Results

A total of 665 patients were identified by ICD-9 codes during the study period, and medical record review disclosed that, in 236 of these children, the IH was located in a cosmetically sensitive facial area (Children's National Medical Center, 187 [79%]; and Virginia Dermatology Practice, 49 [21%]) (Figure 1). Despite exhaustive attempts, only 144 of 236 potential participants (61%) were reached by telephone since their initial evaluation nearly a decade earlier. Thirty of these 144 individuals (21%) completed the surveys and were the participants in this study; 19 (63%) from the initial mailing in 2013 and 11 (37%) from a follow-up mailing in 2014. All 30 respondents completed the SASC-R/Parent and SASC-R/Child surveys, and 29 parents completed the SCI survey. Eleven parents (37%) sent photographs. The concordance rate of the cosmetic impact of the involuted IH was high, with

Table 1. Demographic and Clinical Information of 30 Children With IHs

Characteristic	No. (%)
Sex	
Female	19 (63)
Male	11 (37)
Age, mean (range), y	10.0 (5.4-12.9)
IH location	
Single facial unit	25 (83)
>1 Facial unit	5 (17)
Dominant facial hemangioma location	
Periocular	10 (33)
Nasal	6 (20)
Cheek	5 (17)
Lip or perioral region	4 (13)
Forehead	4 (13)
Ear	1 (3)
Treatment	
Yes	18 (60)
No	12 (40)
Parent's highest educational level	
High school	2 (7)
College	11 (37)
Graduate school	12 (40)
Not reported	5 (17)
Child's problems ^a	
Nonmedical	
ADHD	2 (7)
Borderline ADHD	1 (3)
Learning disability	2 (7)
Anxiety	1 (3)
Trouble decoding social cues	1 (3)
Socially immature	1 (3)
Medical	
Reactive airway disease	2 (7)
Environmental allergies	2 (7)
Seizure disorder	1 (3)
Cholesteatoma	1 (3)
Race	
White	24 (80)
Black	3 (10)
Undeclared	3 (10)
Ethnicity	
Hispanic	1 (3)
Non-Hispanic	29 (97)

Abbreviations: ADHD, attention-deficit/hyperactivity disorder; IHs, infantile hemangiomas.

^a Reported by parents.

all 3 surgeons concurring on 10 of the 11 images. The one dissenting image showed ptosis from an orbital IH and was counted as major per assessment of 2 of the 3 reviewers.

Demographic Data

Demographic data are presented in Table 1. The female to male ratio was approximately 2:1. The mean age was 10.0 years

Table 2. Comparisons of SAS and SCI Survey Scores

Scale	Score, Mean (SD) ^a	P Value ^b
SASC-R/Parent		
FNE		
Sample	15.5 (5.3)	<.001
Normative data	20.8 (6.9)	
Treatment	16.2 (5.8)	.38
No treatment	14.2 (4.1)	
Female	15.5 (5.0)	.99
Male	15.5 (6.0)	
Child	15.6 (1.0)	.15
Parent	17.6 (1.0)	
SAD-New		
Sample	13.1 (4.7)	.004
Normative data	16.0 (5.0)	
Treatment	11.5 (3.8)	.02
No treatment	15.5 (5.1)	
Female	12.6 (5.0)	.53
Male	13.8 (4.3)	
Child	13.1 (0.9)	.69
Parent	13.6 (0.9)	
SCI		
Prosocial Orientation		
Sample	3.96 (0.48)	.50
Normative data	3.89 (0.55)	
Treatment	4.02 (0.56)	.38
No treatment	3.86 (0.31)	
Female	4.04 (0.45)	.21
Male	3.81 (0.52)	
Social Initiative		
Sample	3.81 (0.57)	.07
Normative data	4.03 (0.63)	
Treatment	4.03 (0.55)	.006
No treatment	3.45 (0.43)	
Female	3.88 (0.47)	.47
Male	3.71 (0.72)	

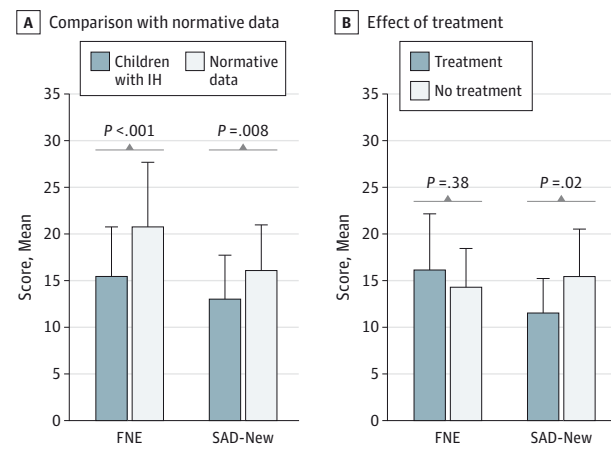
Abbreviations: FNE, Fear of Negative Evaluation Scale; SAD-New, Social Avoidance and Distress in New Situations; SASC-R/Parent, Social Anxiety Scales for Children-Revised completed by parents; SCI, Social Competency Inventory.

^a Higher scores for the FNE and SAD-New indicate worse social anxiety, and lower scores for Prosocial Orientation and Social Initiative domains indicate poorer social competency.

^b P values were determined using paired t tests.

(range, 5.4-12.9 years), and 24 respondents (80%) declared their race as white. Twenty-five children (83%) had a single IH, and the remaining 5 (17%) had multiple IHs, with at least 1 in a cosmetically sensitive area. The periocular region was the most common site (10 patients [33%]), followed by the nose (6 [20%]), cheek (5 [17%]), forehead (4 [13%]), lip or perioral region (4 [13%]), and ear (1 [3%]) locations. Nineteen individuals (63%) received single or dual therapy for their hemangioma, although it was unclear whether that therapy was provided during proliferation. Treatment was prednisolone (10 patients [33%]), and/or surgery (6 [20%]), and/or laser therapy (6 [20%]).

Figure 2. Mean Scores on Social Anxiety Scale for Children-Revised



Higher scores indicate greater social anxiety. A, Participants (sample) with infantile hemangiomas (IHs) did not show greater Fear of Negative Evaluation (FNE) or Social Avoidance and Distress for New Situations (SAD-New).

B, Children with untreated IHs had significantly higher scores for SAD-New compared with those with treated IHs. Error bars indicate SD.

Some parents relayed that their children received diagnoses of psychological or learning concerns, including attention deficit/hyperactivity disorder (2 patients [7%]), anxiety (1 [3%]), mild learning disabilities (2 [7%]), trouble deciphering social cues (1 [3%]), and social immaturity (1 [3%]). Six parents (20%) reported other medical problems (Table 1). This small study did not allow for meaningful statistical analysis of these clinical features. Parents reported their highest level of education as graduate school (12 parents [40%]), college (11 [37%]), high school (2 [7%]), and not reported (5 [17%]). Three parents (10%) reported seeking psychological support before being married ($n = 1$), related to divorce ($n = 1$) and for reasons “unrelated to son or his IH” ($n = 1$).

SASC-R Survey Reliability

All psychiatric subscales of SASC-R showed good reliability (Cronbach $\alpha > 0.70$), except the SAD-General subscale (Cronbach $\alpha = .50$), which remained low (0.57) even after the least correlative item was removed. As such, the SAD-General subscale was not used in the analysis. The results of the t tests are reported in Table 2 and shown in Figure 2.

FNE Survey Results

Participants reported lower FNE scores compared with those of a historic control population indicating less fear of negative evaluation (mean [SD], 15.5 [5.3] vs 20.8 [6.9]; $P < .001$). There was no significant difference in responses between male and female participants ($P = .99$) as well as those with and without prior IH treatment ($P = .38$). The parent and child FNE scores also did not differ significantly ($P = .15$).

SAD-New Survey Results

Participants reported lower SAD-New scores compared with a historic control population indicating normal social functioning (13.1 [4.7] vs 16.0 [5.0]; $P = .004$). There was no sig-

nificant difference in responses between sexes ($P = .53$). However, most children who did not receive IH treatment had significantly higher scores, indicating greater social anxiety, than children who received treatment (15.5 [5.1] vs 11.5 [3.8]; $P = .02$). The parent and child SAD-New scores did not differ significantly ($P = .69$). The parent scores are reported here.

SCI Survey Reliability Results

Both the Prosocial Orientation and the Social Initiative domains of SCI showed high Cronbach α levels (0.88 and 0.76, respectively). These findings indicated good reliability

Prosocial Orientation Domain Survey Results

With results reported as mean (SD), the t test results showed no significant difference in mean Prosocial Orientation domain scores between participants and the corresponding historic controls (3.96 [0.48] vs 3.89 [0.55]; $P = .50$). In addition, no significant differences were determined between females and males (4.04 [0.45] vs 3.81 [0.52]; $P = .21$), or between children with and without prior treatment (4.02 [0.56] vs 3.86 [0.31]; $P = .38$) (Figure 3).

Social Initiative Domain

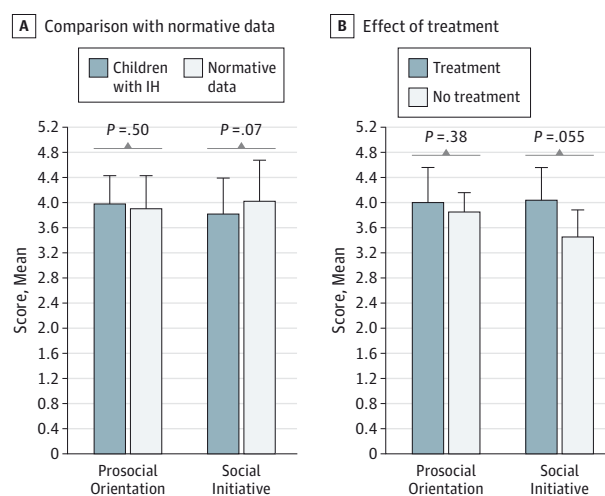
The t test results were marginally significant, indicating a tendency toward lower (poorer) mean Social Initiative domain scores in our sample compared with historic controls (3.81 [0.57] vs 4.03 [0.63]; $P = .07$). Participants without prior IH treatment had significantly lower mean scores compared with those receiving treatment, indicating diminished Social Initiative in the untreated group (3.45 [0.43] vs 4.03 [0.55]; $P = .006$) (Figure 3).

Only 11 participants submitted recent photographs of their child's IH, and the quality was too poor to assess residual skin changes. Four of 11 photographs showed major cosmetic deformities and 7 presented minor deformities. All 4 children with major deformities had received treatment, but only 3 of the 7 with minor deformities had received treatment. Median values for minor vs major deformities for FNE were 22.00 (range, 10-28) vs 13.00 (range, 8-22); for SAD-New, 15.00 (range, 11-16) vs 9.50 (range, 6-12); for Prosocial Orientation, 4.06 (range, 3.47-4.41) vs 4.29 (range, 3.53-4.76); and for Social Initiative, 3.88 (range, 3.25-4.50) vs 4.31 (range, 3.88-5.00). Social anxiety scores were worse for minor vs major deformities; however, the small sample size prevented statistical analysis.

Discussion

Both SASC-R and SCI scores indicated reduced social competency in unfamiliar situations for children not receiving IH treatment compared with those receiving treatment. These results suggest that lack of treatment of IH in cosmetically sensitive areas may decrease social competency and increase social anxiety in new situations for preteen children. Compared with normative data, the SCI scores of all participants with IHs, regardless of treatment status, tended to be lower for the Social Initiative domain, referring to the ability to establish social connections in unfamiliar settings.

Figure 3. Mean Scores on Social Competence Inventory Survey



A, Social Initiative domain scores were similar between participants (sample) and normative data but tended to be worse for participants in the Social Initiative domain. B, Social Initiative domain scores were significantly lower (worse) in children with untreated infantile hemangiomas (IHs) compared with those with treated IHs. Error bars indicate SD.

This study uniquely explored the social anxiety levels and social competency skills of preteen, school-aged children with facial IH. Most prior studies^{3,9} assessing long-term IH effects assessed the parents' response to their child's IH during infancy. Several studies^{5,10} assessed the effect of IHs on children using less-specific health-related quality-of-life questionnaires. Although the investigators concluded that the quality of life of most children is not adversely affected, many lesions were not on the face. Indeed, children who received corticosteroid therapy (suggesting larger or symptomatic IHs) and those with scars tended to have lower pediatric quality-of-life scores and lower physical self-esteem scores than age-matched control children without IHs.⁵

Our study uniquely used 2 validated psychiatric instruments for social anxiety in children instead of the less-specific health-related quality-of-life questionnaires. Prior to the advent of these measures, most studies assessing anxiety used the Revised Children's Manifest Anxiety Scale assessing generalized anxiety instead of the distinctly different social anxiety explored here.¹¹ Children with higher levels of social anxiety report fewer close friendships and a more negative perception of social acceptance and are reported in children rejected or excluded by their peers.¹² Although behavioral conduct and academic competency are not necessarily affected in children with social anxiety, the adverse social phenomena that are affected suggest that facial IHs at risk of increasing a child's social anxiety should be treated during infancy if safe treatment is available.¹²

Social concerns are an important starting point for anxiety in adolescents.¹³ The quality of peer relationships in childhood play a role in healthy emotional development, the development of social skills, and the feeling of personal competence.⁶ Social anxiety can affect interactions with peers

and may lead to several childhood anxiety disorders, including social phobia.

In the absence of contraindications, few clinicians would argue against treating symptomatic IHs during infancy, but treating asymptomatic lesions in cosmetically sensitive facial areas is more controversial. Although reports from the 1950s advocated avoidance of treatment, assuming that all IHs involute nearly completely,¹⁴ we now recognize that most children have cosmetic sequelae of their IHs.¹⁵ The characteristic dimpled orange-peel appearance of skin, fibrofatty changes, and telangiectasias can be deforming. Even lesions smaller than 1 cm may cause devastating deformity when located on the nasal tip or lip, and ulcerated lesions are associated with even worse scarring.¹⁶ Literature on cleft lip and palate^{17,18} reports poorer self-concept and higher anxiety in children with craniofacial anomalies, which supports treatment of such lesions. If involuted IHs in cosmetically sensitive areas affect a child's social comfort or social initiative, then it seems reasonable to treat the lesions during infancy if safe treatment is available.

The participants in this study receiving treatment for their facial IH did not demonstrate higher degrees of social anxiety or lower scores in social competency, although, because they were receiving treatment, they likely had larger facial lesions. These findings suggest that treatment of facial IHs in cosmetically sensitive areas may protect against social concerns later in life.

Nearly 45% of the parents surveyed attended college and another 45% attended graduate school. It is likely that highly educated parents have a greater ability to seek strategies to help their children cope with their IH should it cause social problems. Consequently, our results may be skewed and underestimate the social impact of involuted facial IH in the general population.

The SASC-R scores of parents and children were not significantly different from each other in this study. This finding suggests that the parents' perception of their child's social skills parallels their child's perception.

The Prosocial Orientation domain showed the least variability in participants compared with normative data regardless of treatment, sex, or scarring. This finding suggests that empathy and willingness to console others is not adversely affected by the presence of facial IHs.

The interesting findings of this small pilot study suggest that additional research exploring the social impact of IHs in preteens is necessary. Although the findings reached statisti-

cal significance, this study has limitations, including potential selection bias given that only 21% of potential participants completed the surveys, and may represent a skewed population more affected by their facial IHs and more motivated to respond. We were not able to compare type and duration of treatments given the small sample size and heterogeneity of treatment. In addition, we did not require submission of high-quality facial photographs because we were concerned that doing so would further limit enrollment. Seven of the 11 photographs submitted showed minor residual scars in the children. Although statistical analysis could not be completed on these photographs owing to the small sample size, it is interesting that their median scores, compared with those of children who had major scars, were higher (worse) for both the SAD-New and FNE domains and lower (worse) for Social Initiative. All 4 of the children with major residual scars had received prior treatment compared with only 3 of the 7 with minor cosmetic deformities. Future studies may determine whether the social impact of the residual facial scar is less dependent on its size and more dependent on whether prior treatment is rendered and whether that treatment enhances social confidence regardless of its outcome. Another important consideration is that some preteens undergoing treatment may have recognized the limitations of treatment and learned to not let their facial lesion affect their social competency.

Completing a multi-institutional prospective study with a larger cohort would address many of these study limitations. However, the improved tolerability of propranolol over prednisolone seems to have lowered the threshold for treating cosmetically altering IHs; therefore, completing such a study now would likely be difficult since patients with small lesions in cosmetically sensitive areas often receive treatment.

Conclusions

Preteen children with untreated, involuted facial IHs in cosmetically sensitive areas had higher social anxiety scores and lower social initiative scores compared with preteen children who received treatment for their facial IH. Although this study is limited by a small sample size, these results suggest that treatment should be considered for infants with IHs in cosmetically sensitive areas as it may decrease the risk of social inadequacies during the preteen years.

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Study concept and design: Costa, Haimowitz, Bauman.

Acquisition, analysis, or interpretation of data: All authors.

Drafting of the manuscript: Costa, Haimowitz, Silverman, Bauman.

Critical revision of the manuscript for important intellectual content: Costa, Haimowitz, Cheng, Wang, Bauman.

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