

The pediatric patient at a maxillofacial service – eye prosthesis

O paciente pediátrico em um serviço de prótese bucomaxilofacial – prótese ocular

Beatriz Silva Câmara Mattos*
Maria Cecília Montagna**
Clemente da Silva Fernandes***
Antonio Carlos Lorenz Sabóia**

ABSTRACT: Congenital absence or loss of the ocular globe during childhood causes psycho-social and cosmetic disorders and compromise the normal development of the orbital region. The literature relating to congenital or acquired etiology, due to trauma or disease, demonstrates the necessity of prevention and early detection in order to minimize the sequelae and disturbances in orbital growth. Installation of an eye prosthesis is essential to the rehabilitation process, so as to produce satisfactory development of the region. In order to characterize a profile of the child patient with this condition, a survey was carried out at the Prosthetic Eye sector, Out-patient Clinic, Discipline of Maxillofacial Prosthodontics, School of Dentistry, University of São Paulo (FOUSP), during the period from 1988 to 2003. The 124 (14.02%) patients within the age group of 0-13 years registered for ocular prosthesis were divided into a first group of 64 patients (51.62%) with 0-7 years, and a second group of 60 patients (48.38%) with 8-13 years. Fifty nine were girls and 65 were boys. No statistical significance was observed regarding the distribution of genders in the two analyzed age groups ($p = 0.069$). However, there was statistical significance at the level of 0.01 in relation to etiology, with higher prevalence of congenital and pathological disturbances in the younger group and traumatic occurrences in the older group. The etiology also presented variation according to the gender, at the significance level of 0.05, where girls presented three times less trauma than boys in the older age group. The necessity of prosthetic ocular repair was evenly distributed along the childhood period and the eye losses that required prosthetic treatment equally affected both genders. However, the etiology of eye loss varied according to the considered gender and age bracket.

DESCRIPTORS: Ocular prosthesis; Anophthalmos; Eye; Eye injuries; Pediatrics.

RESUMO: A ausência congênita ou a perda do bulbo ocular na infância acarretam distúrbios estéticos e psico-sociais e comprometem o desenvolvimento normal da região orbitária. A literatura relacionada à etiologia congênita ou adquirida, devido a trauma ou doença, demonstra a necessidade da prevenção e da detecção precoce para minimizar as seqüelas e os distúrbios de crescimento orbital. A reabilitação implica a instalação de uma prótese ocular visando favorecer o desenvolvimento harmonioso da região. Com o objetivo de caracterizar o perfil do paciente infantil foi realizado um levantamento junto ao Setor de Prótese Ocular do Ambulatório da Disciplina de Prótese Bucocomaxilofacial da FOUSP abrangendo o período de 1988 a 2003. As 124 (14,02%) crianças registradas encontravam-se distribuídas em 64 (51,62%) na faixa etária de 0 a 7 anos e 60 (48,38%) na de 8 a 13 anos de idade, sendo 59 meninas e 65 meninos. A análise estatística revelou que a variação das ocorrências dos gêneros masculino e feminino nas faixas etárias não é significativa ($p = 0.069$). Entretanto, a etiologia da perda variou segundo as faixas etárias consideradas, com um nível de significância de 0,01, apresentando uma maior prevalência de distúrbios congênitos e patológicos na faixa etária mais jovem e trauma na mais velha. A etiologia alterou-se de acordo com os gêneros com uma significância de 0,05, tendo as meninas apresentado três vezes menos trauma que os meninos na faixa etária mais velha. A necessidade de tratamento protético apresentou-se igualmente distribuída ao longo da infância e as perdas oculares que implicam reabilitação protética ocorreram igualmente nos gêneros masculino e feminino. Entretanto, a etiologia da perda ocular variou em função do gênero e da faixa etária considerada.

DESCRIPTORIOS: Prótese ocular; Anoftalmia; Olho; Traumatismos oculares; Pediatria.

INTRODUCTION

The loss of the ocular globe during childhood, due to congenital, traumatic or pathological aetiologies, affects the growth and development of the

orbital area, which may result in hypoplasia and facial asymmetry, and represents an additional factor of aesthetic and psychological misbalance.

* Associate Professor; **PhDs, Professors; ***Master's Degree Student – Department of Maxillofacial Surgery, Prostheses and Traumatology, School of Dentistry, University of São Paulo.

Rahi *et al.*¹¹ (1995) mention that the etiology of congenital anomalies includes chromosomal and genetic disorders, where the great majority of cases presents a recessive autosomal pattern. According to Levin⁸ (2003), microphthalmia can be a primary ocular disorder or a secondary manifestation of a craniofacial malformation, as lateral facial dysplasia. It can also be part of a wide variety of syndromic multi-systemic disorders. At the same time, viral infections such as German measles, measles and toxoplasmosis during gestation, as well as the use of teratogenic drugs during that period, can lead to a disturbance in the development of anatomical structures, resulting in congenital cataract and hypoplasia of the ocular globe. Wallace *et al.*¹⁸ (1998) consider that measles complications or secondary glaucoma due to congenital German measles can also provoke ocular globe enucleation.

Children that present congenital anophthalmia usually develop a small orbita, hypoplastic soft tissues and shortening of the eyelid rima. Sá Lima *et al.*¹³ (2002) remind that the rehabilitation process of these cases has been traditionally performed with the previous use of expander prostheses in acrylic resin, with periodic changes for successive increases, until an appropriate dimension and volume are obtained to allow a prosthetic iris incorporation. Schittkowski *et al.*¹⁴ (2003) reported the inflatable hydrogel use for dilation of the soft tissues and stimulation of the orbital bone tissues. Considering the rehabilitation aspects, Heher *et al.*⁵ (1998) reported that dermis-fat grafts performed in 16 children with microphthalmia or anophthalmia, used concomitantly with prosthetic conformers, presented a good progression, allowing orbital growth. According to these authors, this procedure is used in order to expand the eyelid rima, and to provide a deepened fornix and posterior area of the orbital cavity, creating space for a possible implant. While the prosthetic conformers suffer successive increases, they concomitantly stimulate tissue and orbital growth, providing a potentially improved symmetry between the healthy and non-healthy sides.

Ocular globe injuries are the main cause of ocular morbidity, potentially leading to loss of structures and requiring the installation of an ocular prosthesis. Epidemiological studies on the occurrence of ocular injuries due to perforating-contusive objects, domestic accidents, automobile accidents and urban violence identify possible risk factors and suggest valuable preventive measures^{6,15,16,20}. Cole, Smerdon² (1988), through a research about

perforating injuries of the ocular globe, registered 375 patient cases, among which 78 (20.8%) were observed in children with less than 14 years of age. Hemady⁶ (1994), in a one-year prospective study of ocular traumas in an urban population, compiled 530 patient cases, among which 110 were patients with less than 18 years of age (21%), 85 (77%) were males and 25 (23%) were females. A research about the occurrence of traumas to the ocular globe due to personal violence accomplished by Zagebaum *et al.*²⁰ (1993) revealed that, of the 277 registered patients, 41 (15%) were children, with an average age of 10 years, of which 34 suffered aggressions from older children and 7 suffered aggressions from adults.

Rahi *et al.*¹¹ (1995) reported a survey accomplished in India with 1,318 children with serious visual damage and blindness, with ages ranging from 3 to 15 years. Of these, 764 (58%) were males, 53 (4%) had less than 5 years, 527 (40%) had ages ranging from 6 to 10 years while 738 (56%) had ages varying from 11 to 15 years. The most commonly observed causes were cornea diseases followed by alterations of the ocular globe such as anophthalmia, microphthalmia and other diseases of ignored etiology.

However, according to Heher *et al.*⁵ (1998), ocular tumors such as retinoblastoma are the major causes of ocular globe enucleation during first childhood. In a study with childhood cancer series, the retinoblastoma has been ranked second in occurrence, corresponding to 17.1% of all types of cancer occurring in infants¹⁹. Considered the most common primary intraocular malignancy of childhood, this neuroblastic tumor may be unilateral or bilateral¹⁰. Retinoblastoma incidence in Brazil is approximately 1:20,000 live births, with an incidence peak at 18 months of age¹⁷. In a retrospective study carried out at the Cancer Hospital of São Paulo, Department of Pediatrics, Rodrigues *et al.*¹² (2004) observed that out of 327 patients, 52.3% were male and 47.7% were female, with a 1.1:1 male/female ratio. Desjardins *et al.*³ (2000), reviewing a series of 153 children treated for retinoblastoma, found out that 56% of them were girls and 44% were boys.

Vieira¹⁷ (2003) considers that eye enucleation has been the method of choice in unilateral cases, suggesting that radiotherapy and chemotherapy be used in bilateral cases to preserve vision possibility at least in one eye. Desjardins *et al.*³ (2000) report that among 76 cases of unilateral retinoblastoma, 56 eyes were enucleated and 20 were

TABLE 1 - Distribution of children in the 0 to 7 years and 8 to 13 years age groups according to gender and etiology of ocular globe loss (number and percentage).

Age group n (%)	Gender		χ^2	Etiology		χ^2
	Male n (%)	Female n (%)	sig.	Congenital/Pathological n (%)	Traumatic n (%)	sig.
0-7 – 64 (51.62)	28 (43.75)	36 (56.25)	n.s.	56 (87.50)	8 (12.50)	0.01
8-13 – 60 (48.38)	37 (61.67)	23 (38.33)		22 (36.67)	38 (63.33)	

sig.: significance; n.s.: not significant.

treated conservatively, whereas among 154 eyes with bilateral retinoblastoma, 48 were enucleated and 106 were treated conservatively (49 by external beam radiation). Jackson *et al.*⁷ (1996) remind that children receiving radiotherapy in the orbital area frequently develop craniofacial deformities affecting not only the orbit but also the cranium, maxilla and mandible. Evaluating facial deformities resulting from radiotherapy in the orbital area during childhood, Fountain *et al.*⁴ (1999) observed that the orbital volume on the affected side was reduced from 21.1% to 55.8%, with an average of 37.8%. According to Aihara *et al.*¹ (1988), peri-orbital deformity due to radiotherapy after ocular globe enucleation hinders the installation of an ocular prosthesis, compromising the aesthetic result of rehabilitation.

In an epidemiological survey carried at the Maxillofacial Prosthetic Service, School of Dentistry, University of São Paulo, from January 1976 to December 1985, Mattos⁹ (1987) reported 558 patient records of loss of and atrophies in the ocular globe, 72 cases (13.1%) with ages ranging from 0 to 10 years, with no significant statistical differences between genders or between the right and left sides.

The present study was undertaken to identify the service demands of the infantile population, as well as to characterize the infantile patient profile regarding age, etiology and gender of those patients attended at the Maxillofacial Prosthetic Service, School of Dentistry, University of São Paulo.

MATERIAL AND METHOD

A survey was carried out based on the health charts of patients attended at the Maxillofacial Prosthetic Service, School of Dentistry, University of São Paulo, with ocular globe loss and registered for ocular prosthesis, from 1988 to 2003. The data were collected in specifically elaborated spreadsheets, considering age groups from 0 to 7 years and from 8 to 13 years. The variables observed

TABLE 2 - Distribution of the etiologies of the ocular globe loss according to male or female genders (number and percentage).

Gender n (%)	Etiology		χ^2
	Congenital/ Pathological n (%)	Traumatic n (%)	sig.
Male 65 (52.42)	33 (50.77)	32 (49.23)	0.05
Female 59 (47.58)	45 (76.27)	14 (23.73)	

sig.: significance.

were gender and etiology of loss of the ocular globe. The data were submitted to chi-square statistical analysis.

RESULTS

The 884 patient registers from the Section of Ocular Prosthesis pertaining to the period included in this work revealed a number of 124 children (14.02%): 64 (51.62%) in the group from 0 to 7 years and 60 (48.38%) in the group from 8 to 13 years.

The distribution and percentages of occurrences that entailed different modalities of ocular prosthesis confection were tabulated according to gender and etiology of loss of the ocular globe for the two age groups (Table 1). The congenital and pathological etiologies were grouped for better interpretation of the results.

The distribution of treatment occurrences according to etiology in the male and female genders is presented in Table 2.

The statistical analysis revealed that the variation of occurrences in the male and female genders in the studied ages was not significant ($p = 0.069$). However, the etiology of loss of the ocular globe varied according to the age groups, with a highly significant level of 0.01 ($p = 0.000$). At the same time, the etiologies varied according to gender, presenting a significance level of 0.05 ($p = 0.011$).

DISCUSSION

Mattos⁹ (1987), in a previous survey of 10 years in the same Maxillofacial Prosthetic Service, observed that 13.1% of the patients were in the age group from 0 to 10 years. This value is very close to the 14.02% of the present study, which indicates stability in the afflux of infantile patients for confection of ocular prostheses.

The balance between the age groups of 0-7 and 8-13 years suggests that the search for prosthetic rehabilitation has been made at an early stage. The importance of rebuilding the content volume of the orbital cavity for the correct growth and development of the anatomical structures is well-established⁵. Children with congenital anophthalmia or microphthalmia demand a two stage treatment to recover orbital and eyelid symmetry through expansive or dilator prostheses^{5,13} and to avoid additional disturbance in the development of the orbital area. These goals might be accomplished by means of successive eye prostheses, providing the children with a normal appearance⁸. The results of this study indicate a concern in rescuing possible orbital atrophies congenitally established as soon as possible. The higher occurrence of pathological etiology in the age group 0 to 7 years may also be explained by the many cases of retinoblastoma¹⁹, the major cause of ocular globe enucleation in first childhood⁵, not specified in this study but included in the pathological category. Melamud *et al.*¹⁰ (2006) found that the mean age at diagnosis was 12 months for bilateral tumors and 24 months for unilateral tumors; Rodrigues *et al.*¹² (2004) observed that 53% of diagnoses occurred before 2 years of age, 44% between 2 and 6 years and only 3% after six years. These age groups at diagnosis justify the higher occurrence of pathological etiology in the 0 to 7 years group in the present study, allowing the conclusion that children have been led to prosthetic rehabilitation at a time nearby the time of occurrence of the lesion. Thus, prevention of the hypodevelopment of the orbital area after enucleation of the ocular globe during childhood using conventional ocular prostheses^{5,18} has been done.

The statistical analysis indicated that a variation in patients of the male and female genders in the considered age groups was not significant ($p = 0.069$), although existent, indicating that boys and girls sought treatment along childhood in the same way. The significant variation in etiologies in the age groups, with prevalence of the traumatic

etiology in the 8 to the 13 years group, was expected and also indicates that these children were led to the service at a time nearby the time of occurrence of the lesion. It is important to remind that the exposure to the risk of trauma increases along the period of age considered in the present study, thus justifying the significance level of 0.01. The observation that 20.8% of perforating injuries of the ocular globe occurred in children with less than 14 years of age² is thus confirmed by the results of the present study. A previous study, developed in the same maxillofacial service but considering different age groups, also pointed out a significant increase of loss of the ocular globe due to trauma during adolescence, with the 11 to 20 years group presenting the highest percentage of loss (18.8%), which corresponded to traumatic etiology⁹.

The observation that the etiologies varied according to gender, with a significant level of 0.05, should consider the fact that the male gender presented a quite balanced distribution between the etiologies, while in the female gender the occurrence of the traumatic etiology was 1/3 smaller in the 8 to 13 years age group. The observation that 21% of ocular trauma in patients with less than 18 years of age, comprising 77% of males and 23% of females⁶ is thus confirmed by the results of the present study. We speculated that girls, considering their social behavior, might not engage in risk activities as much as boys do, thus being less subjected to traumas in the ocular area. The expressive participation of the traumatic etiology, responsible for 37.09% of the total number of ocular prosthesis cases in childhood observed in the present study, reinforces the importance of effective measures in order to prevent ocular globe trauma. The predominance of congenital and pathological etiology in females was probably occasional since there is no evidence in the literature justifying this result. Yet, as mentioned before, the present study did not specify the disease, but there were many cases of enucleation due to retinoblastoma, a tumor equally affecting boys and girls^{3,12}.

The use of conventional ocular prostheses during childhood entails periodic changes with successive increases in order to accompany the expansion of the anophthalmic cavity, and it is the only way to aesthetically rebuild the anophthalmic socket. It is necessary to objectively determine a time-table of ocular prosthesis change along the whole period of cranio-facial growth in order to minimize the possible discrepancy between the compromised and the healthy sides, thus contributing to balance

and harmony of the facial development. Hence, the installation of an ocular prosthesis still during first childhood adds an inestimable psychological and social contribution to the physical benefit in the patient's global rehabilitation.

CONCLUSIONS

1. Variation of the male and female genders in the different age groups was not significant, indicating that boys and girls sought treatment along childhood in the same way.

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