

Orbital Exenteration: a series of cases

Exenteração orbitária: série de casos

Gabriel de Almeida Ferreira¹, Natalia Mussi¹, Roberta Lilian Fernandes de Sousa Meneghim², José Vicente Tagliarini², Mariângela Esther Alencar Marques³, Silvana Artioli Schellini²

ABSTRACT

Objective: To describe causes of orbital exenteration in a Brazilian tertiary hospital. **Methods:** A retrospective study was done, involving patients submitted to orbital exenteration at the Clinical Hospital of Botucatu Medical School, between the years of 1993 to 2016. The surgeries have been performed under general anesthesia, by a multidisciplinary team, composed by ophthalmologists, otolaryngologists and head and neck surgeons. **Results:** Fourteen cases of orbital exenteration occurred in the period of the study, with a mean age of 63.36 ± 13.18 years and nine were men (64.3%). All exenteration were due to malignant tumors, being more frequent the squamous cell carcinoma (7 cases - 50.0%). The most common primary sites were the eyelids (50.0%) followed by the conjunctiva (28.6%). The majority of the surgeries was extended exenteration type (57.1%) and most of the reconstructions was made by spontaneous granulation (64.3%). The survivor rate in 1 year was 78,6% and in 5 years was 71.4%. **Conclusion:** The main cause of orbital exenteration was squamous cell carcinoma and the most frequent primary site was the eyelids. Extended exenteration was necessary for the majority of cases, most of them with free margins.

Keywords: Orbit exenteration; Orbital neoplasms; Squamous cell carcinoma; Frequency of occurrence

RESUMO

Objetivo: Descrever os casos de exenteração orbitária de um hospital terciário brasileiro. **Métodos:** Estudo retrospectivo, envolvendo pacientes submetidos à exenteração orbitária no Hospital das Clínicas da Faculdade de Medicina de Botucatu, entre os anos de 1993 a 2016. As cirurgias foram realizadas sob anestesia geral, por equipe multidisciplinar composta por oftalmologistas, otorrinolaringologistas e cirurgiões de cabeça e pescoço. **Resultados:** Foram estudados 14 casos de exenteração orbitária, com média de idade de $63,36 \pm 13,18$ anos e nove homens (64,3%). Todas cirurgias foram realizadas para tratamento de tumores malignos, sendo mais frequente o carcinoma espinocelular (7 casos - 50,0%). Os sítios primários mais frequentes foram as pálpebras (50,0%), seguida pela conjuntiva (28,6%). A maioria das cirurgias foram do tipo exenteração estendida (57,1%), com cicatrização por granulação espontânea (64,3%). A sobrevida em 1 ano foi de 78,6% e em 5 anos de 71,4%. **Conclusão:** O carcinoma espinocelular foi a principal causa de indicação de exenteração orbitaria, sendo as pálpebras o sítio primário mais frequente. O procedimento mais realizado foi a exenteração estendida, com a grande maioria alcançando margens livres.

Descritores: Exenteração orbitária; Neoplasias orbitárias; Carcinoma espinocelular; Frequência de ocorrência

¹ Residency Program in Ophthalmology, Faculdade de Medicina de Botucatu, Universidade Estadual Paulista "Júlio de Mesquita Filho" Botucatu, SP, Brazil.

² Department of Ophthalmology, Otorhinolaryngology and Head and Neck Surgery, Faculdade de Medicina de Botucatu, Universidade Estadual Paulista "Júlio de Mesquita Filho" Botucatu, SP, Brazil.

³ Department of Pathology, Faculdade de Medicina de Botucatu, Universidade Estadual Paulista "Júlio de Mesquita Filho" Botucatu, SP, Brazil.

Institution: Hospital das Clínicas, Faculdade de Medicina de Botucatu, Universidade Estadual Paulista "Júlio de Mesquita Filho" Botucatu, SP, Brazil.

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INTRODUCTION

The orbital exenteration is a radical procedure that consists in removing the content of orbit including the eye bulb, orbital fat, periocular muscles, conjunctival fornix and all or part of the eyelid.⁽¹⁾

There are three types of exenteration: total, when the removal is of all content of orbit, with or without removal of the eyelids; subtotal, when the removal of the orbital content is partial, but with sacrifice ocular bulb; and extended exenteration, when adjacent bone walls or paranasal sinuses are also removed.⁽²⁾

The classic indication for this procedure is malignant tumors, and it may be required due to orbital invasion secondary to malignant neoplasms of ocular annexes like the eyelids,¹⁻³ conjunctival lesions,⁴ primary orbital tumours,⁵ and less commonly benign lesions which have intractable pain, inflammatory disease or pseudotumors.¹

After the exenteration it is necessary to reconstruct the orbital cavity, with the possibility of using the temporal muscle transposition or skin flaps in the region adjacent to coat the orbital cavity or wait for healing by spontaneous granulation.¹

Exenteration studies are scarce in the literature, especially in the Brazilian population, with the objective of this study being to describe the cases of exenteration of a Brazilian tertiary hospital.

METHODS

This is a retrospective study involving patients undergoing orbital exenteration at Hospital das Clínicas da Faculdade de Medicina de Botucatu (HC-FMB-UNESP), between the years of 1993 and 2015. The study was approved by the Ethics Committee of the institution.

The data was recovered from the electronic medical record of the patient, being important age, gender, diagnosis, previous treatments, date of surgery, surgical technique, method of reconstruction, result of pathology and evaluation of surgical margins.

The surgeries were performed under general anesthesia by a multidisciplinary team comprising ophthalmologists, otolaryngologists and head and neck surgeons.

The data was transferred to the Microsoft Excel 2010 spreadsheet, and analyzed by the program IBM Statistical Package for the Social Sciences (SPSS) version 20. Continuous variables are expressed by mean values and standard deviation, and the qualitative ones by frequency and absolute number. We considered $p < 0.05$ as significant.

RESULTS

In the period of evaluation 14 exenterations were carried out in 14 patients, being the average age of 69.36 ± 13.18 years (ranging from 45 and 88 years). The study population comprised nine men (64.3%) and five women (table 1).

All exenterations carried out were for treatment of malignant tumors. The histopathological analysis showed five different types of tumors, and squamous cell carcinoma (SCC) was the most frequent with 7 cases (50.0%), followed by three cases of esclerodermiform basal cell carcinoma (BCC) and two melanomas (table 2). The melanomas were the only injuries that did not derive from the annexes, but from the eye itself.

Table 1
Characteristics of patients undergoing orbital exenteration

Characteristics		n (%)
Gender	Male	9 (64.3)
	Female	5 (35.7)
Type of exenteration	Extended	8 (57.1)
	Total	5 (35.7)
	Subtotal	1 (7.1)
Laterality	Left	7 (50.0)
	Right	7 (50.0)
Method of reconstruction	Granulation	9 (64.3)
	Skin flaps	5 (35.7)
Free surgical margins	Yes	13 (92.9)
	No	1 (7.1)

Table 2
Distribution of exenteration cases according to the pathological diagnosis

Anatomicopathological	n (%)
Squamous Cell Carcinoma	7 (50.0)
Basal cell sclerodermiform carcinoma	3 (21.4)
Melanoma	2 (14.3)
Adenocystic carcinoma	1 (7.1)
Sebaceous carcinoma of eyelids	1 (7.1)

The average age of patients with SCC was 65.86 ± 14.40 , and with BCC was 72.00 ± 4.58 years, that is, tumors that have resulted in orbital exenterations took place mainly in the elderly.

The primary sites of tumors are listed in table 3, with the most frequent being the eyelids (seven cases), followed by conjunctiva (four cases). In relation to the surgical margins after exenteration, 13 cases (92.9%) presented free margins in the pathological study (table 1).

Table 3
Distribution of cases of orbital exenteration according to the primary tumor site

Primary site	n (%)
Eyelids	7 (50.0)
Conjunctiva	4 (28.6)
Lacrimal pathway	2 (14.3)
Maxilla	1 (7.1)

Half of the patients (seven cases) had undergone previous treatment, with previous surgical excision being the most frequently modality, isolated, associated to radiation therapy or to the use of 5-Fluoracil, corresponding respectively to four, one and one case (table 4).

The most accomplished type of surgery was the extended exenteration (seven procedures), followed by total exenteration (five procedures), and one case of subtotal exenteration. Regarding laterality, there was no difference between the sides, with seven cases on the left (50.0%) (Table 1).

Table 4
Frequency of previous treatments performed
in patients with tumors that required
orbital exenteration

Previous treatment	n (%)
No previous treatment	7 (50.0)
Surgical excision	4 (28.6)
Surgical excision + 5-Fluoracil	1 (7.1)
Surgical excision + Radiation therapy	1 (7.1)
Radiation therapy	1 (7.1)

The most widely used method of reconstruction was granulation by secondary intention (nine cases), with the remaining five cases being performed by flap rotation (Table 1).

Of the patients studied, four missed follow-up during treatment, so there is no information regarding the time of follow-up and outcome.

In the remaining patients, the time of follow-up after surgery was 35.36 ± 35.10 months (ranging from 0.00 to 90.63 months). There was a total of five deaths during the follow-up period, with a survival rate in 1 year of 78.6%, and of 71.4% in 5 years. The deaths were secondary to a case of adenocystic carcinoma, two of melanoma, one of BCC and one of SCC, however unrelated to the type of tumor ($p > 0.005$). In addition, these patients registered a case of recurrence and one of metastasis, being a case of adenocystic carcinoma and one of melanoma, respectively.

DISCUSSION

The purpose of this study was to show the profile of individuals who suffer orbital exenteration, since there are few data in the literature on the subject, especially in the Brazilian population.

As the orbital exenteration is a mutilating procedure carried out in cases of advanced disease, their prescriptions are limited. In our study, all prescriptions were due to malignant tumors. The prescription can also be for benign tumors or pseudotumors, but always with a greater frequency for malignant neoplasms,⁶ as in Manchester, where among 69 exenterations, 92.7% were due to malignant neoplasms.¹ In another study involving 16 exenterations, all were due to malignant tumors.⁷ Currently, more conservative treatments for SCC and BCC, as local excisions, indicate similar tumor control.³ The search for alternative treatments is probably due to the mutilation induced by exenteration, reflecting on the quality of life compared to the general population.⁸

Considering the subtypes of malignant tumors that most require this type of surgical approach, literature studies differ between SCC and BCC,^{1,7,9} with the majority pointing to the BCC as the most common subtype¹⁰⁻¹² However, squamous cell tumor corresponded to 50.0% of our cases, finding similar to the Australian survey showing the SCC as responsible for 48.0% of exenterations carried out⁹, and another Brazilian study also showing SCC in 54.2% of cases.¹³ BCC is approximately 87% of palpebral tumors¹⁴, and although the SCC is less common than the BCC, it is a more aggressive subtype that may be present with early perineural invasion, with a greater chance of orbital invasion and faster than the BCC. In our study there was a trend of SCC

cases being operated in younger patients, but without statistical significance, perhaps due to the reduced sample. However, another study showed this finding.³

Usually this type of procedure is performed by ophthalmologists, head and neck surgeons, otolaryngologists, plastic surgeons, among others. The exenterations performed by ophthalmologists tend to be of the total or subtotal type, with greater tissue preservation.⁹ In our study, most exenterations performed were of the extended type (57.1%), which can be explained by the fact that we work in a multidisciplinary team.

In addition, another difference is that ophthalmologists opt for healing by granulation, but non-ophthalmologists use other techniques such as temporal muscle flap, skin graft of total or partial thickness, derma-fat graft, among others.¹⁰

The need for adjuvant treatment after surgery like radiotherapy influences the type of reconstruction because, if required, it is necessary to reconstruct it with flaps.¹² Healing by second intention (granulation) is more time consuming and does not allow the completion of radiation therapy. However, it allows early identification of recurrence and a more uniform color of the cavity. The use of flaps and grafts, besides allowing early radiotherapy, leads to a faster healing, with the disadvantage of complicating the diagnosis of recurrences and sometimes complicating the adaptation of prostheses. In our study, 64.3% of cases were reconstructed by second intention healing.

All of our patients are referred for facial prosthesis adaptation after surgery, which can give a better appearance and improve the quality of life.

During the period of study, among patients without tracking loss, 5 deaths were evidenced, with a 1 year survival of 78.6%, and of 71.4% in 5 years. There was no statistical difference between the types of tumors, probably because of the small sample. However, the two cases of melanoma have evolved to death, one of the seven cases of SCC (14.3%), and one of the three cases (33.3%) of BCC. These findings are similar to a study of Massachusetts who found a survival in 1 year of 72%, and a higher mortality in melanomas (85.7%).¹⁵

Among the limitations to our study are having a retrospective design and presenting a small number of patients, possibly justified by the low performance of this type of procedure, making it difficult to have a prospective study with the largest number of patients. Another limitation was the loss to follow-up of four patients, which could add information on survival and time of follow-up.

CONCLUSION

The exenteration indication of a Brazilian tertiary hospital took place predominantly with SCC, and the main primary site was located on the eyelids. The most accomplished procedure was the extended exenteration, with the great majority reaching free margins.

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Corresponding Author:

Gabriel de Almeida Ferreira

Av. Prof. Mário Rubens Guimarães Montenegro, s/n

Bairro: UNESP - Campus de Botucatu18618687 - Botucatu, SP

Phone: (14) 3811-6256