Cholesterol Granuloma of the Orbit. Report of Cases and Analysis of Controversial Treatment

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Abstract Cholesterol granuloma of the orbit is a rare entity originated from a foreign body reaction against cholesterol crystal. Its pathogenesis still remains unclear, while controversy is still present concerning its management. In the present report, two new cases of cholesterol granuloma of the orbit are presented and the authors focus on treatment considerations.

Keywords Cholesterol granuloma orbit · Surgical treatment

Introduction

Cholesterol granuloma of the orbit is a rare entity caused by a foreign body reaction against cholesterol crystals. It has been reported in several locations such as the peritoneum, lungs, breast, lymph nodes, kidney and testis [1, 2]. Within the head and neck location, it has been mostly reported in association with bony structures such as the mastoid antrum and air cells of the temporal bone. It has also been referred in the jaw, nasal sinuses and the base of

the skull [3]. Within the orbital region it has been usually reported in the upper lateral site, of frontal bone in the region of the lacrimal fossa.

In relation to its pathogenesis several hypotheses have been postulated, although no definitive event–effect relationship has been established. Controversy is still present concerning its management. This study was approved by the Institutional Review Board of the University Hospital La Princesa, Madrid.

Report of Cases

Case 1

A 47-year-old man presented with progressive left proptosis without diplopia nor sensorial alterations for 3 years. Physical examination showed proptosis of the left ocular globe, without disturbances in extrinsic ocular movements (Fig. 1). CT-scan and Magnetic Resonance Imaging (MRI) showed an erosive $3.2 \times 2.1 \times 2.8$ cm extraconal well-defined polylobulated mass in the superolateral aspect of the left orbit (Fig. 2A, B) was observed. Close contact with upper and lateral rectus muscles was also noted. An important thickness of the upper–lateral orbital wall was observed. In T2 stage, the lesion was predominantly hyperintense and heterogeneous with a thick hypo-intense ring around it, in relation to a surrounding fibrous wall. A Fine Needle Aspiration Cytology (FNAC) was then performed with the result of multiple yellowish bi-refringent prismatic crystals. No foreign body giant cells were observed. Clinical diagnosis was cholesterol granuloma of the orbit. The patient underwent complete surgical excision of the lesion by means of a bicoronal approach (Fig. 3). Curettage of the upper and lateral orbital wall was also performed by careful

Both authors have contributed equally to the development of the present manuscript and must be cited together with equal importance.

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Fig. 1 Clinical view of the patient at time of diagnosis. Note the evident left proptosis

Fig. 3 Intraoperative view of the lesion following bicoronal approach

Fig. 2 Patient no 1. **A** MRI **B** CT-scan. Note the extraconal well-defined polylobulated mass in the superolateral aspect of the left orbit with erosion of the lateral wall of the orbit

drilling of the bone. No complications were observed following the intervention. Complete disappearance of proptosis was noted during the immediate post-operative period. No relapse of the lesion has been observed 12 months following surgery. Histologic study revealed cholesterol crystals and hemosiderin deposits (Fig. 4A).

Fig. 4 Histological examination. Note disposition of cholesterol crystals and hemosiderin deposits. **A** Patient no 1. **B** Patient no 2

Case 2

A 35-year-old man presented with left diplopia for two months. Physical examination showed severe proptosis of the left ocular globe with alteration of the extrinsic ocular mobility. A CT-scan showed a $2.0 \times 1.2 \times 1.3$ cm left

supraorbital mass in the upper aspect of the left orbit with erosion of the roof of the orbit. Inferior displacement of the ocular globe and intraconal contents was also observed. A simple biopsy of the lesion was initially performed. Histologic examination revealed inflammatory material with abundant cholesterol crystals and macrophages with hemosiderin, several lymphoid cells and scarce fibrous areas (Fig. 4B). Other amorphous and necrotic areas were also observed. Neither epithelial rests nor eosinophils were noted. Further complete surgical excision of the lesion by means of a bicoronal approach was performed. Curettage of the upper orbital wall was performed by careful drilling of the bone. Complete disappearance of proptosis was noted in the immediate post-operative period. No relapse of the lesion has been observed 11 years following surgery.

Discussion

Several hypotheses have been postulated in relation to the pathogenesis of cholesterol granuloma. The most accepted one is the presence of a non-absorbed hematoma from an episode of mucosal bleeding, resulting in formation of cholesterol crystals and a foreign body chronic inflammatory reaction. Within the middle ear, obstruction of air draining may result in edema and hemorrhage and subsequent granuloma formation [4]. However, other explanation such as local tissue breakdown has been reported to play an important role as source of cholesterol in the petrous sinus region [4]. In the frontal sinus, cholesterol granuloma may be caused by disturbance of air drainage by middle nose polyps [5]. Some other authors have also reported a possible traumatic event over the frontal region with hematoma formation and retention of blood coagulated in the frontal sinus [6–8]. Cholesterol granuloma of the orbit has been less frequently associated with a previous trauma [6]. In the present two cases, orbital cholesterol granulomas were not associated with previous trauma. Thus, pathogenesis of cholesterol granuloma still remains unclear and more histologic studies are necessary to improve our knowledge about its origin.

Clinically, proptosis has been reported to be the most common presenting symptom, as in our two cases. Peri-orbital pain and ptosis have also been reported [6]. Compression of the globe and extrinsic ocular muscles may be noticed by an important restriction in ocular movements. Masses are usually located in the superolateral aspect of the orbit and thus inferomedial displacement of the globe and restriction of the superotemporal ocular movement should be expected. This condition may be evident in physical examination and imaging techniques. From this site, masses may invade other neighbour regions, such as the orbital apex, orbital roof, lateral orbital wall and greater sphenoid

wing. Frontal, ethmoid, sphenoid and cavernous sinuses may be also affected by large masses. Although infrequent, reduced visual acuity has been reported in relation to induced astigmatism or choroid folds involving the macula [7].

Due to the absence of epithelial elements [8], cholesterol granuloma of the orbit rarely recur. Consequently, some authors [9] have argued that complete extirpation of the adjacent bone is not necessary. However, recurrences have been reported in relation to the absence of enough bone drilling [6]. This is particularly true if tiny remnants of the original tumor still persist in the bony base. This situation advocates for a complete resection of the mass and a generous bone drilling.

Resection of a wide margin of surrounding bone has not been already proven to be necessary, moreover if the delicate anatomy of structures in the vicinity of the orbit is considered. However, some severe orbitofrontal cases with great deformity of the skull bone and cosmetic problems may require wide approaches with precise cystic wall removal and reconstructive techniques. Although rare, extension in the temporal and anterior cranial fossa has been reported [10]. It may be explained by the ability of the tumor to erode bone and invade other areas. Histopathologic findings of bone necrosis may be present in these extended cases. This condition would make desirable a generous resection of necrotic bone and not only a simple curettage of the lining. Thus, large masses extending into surrounding bone, with clinical and radiological findings of osseous invasion, should be managed more aggressively, with extirpation of necrotic bone of the vicinity and further reconstruction with non-vascularized or vascularized bone graft if needed.

The decision to adopt aggressive treatment measures shall be based on proper clinical and radiological assessment of the individual lesion, as in the present report.

Conflict of interest None.

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