



Case Report

Uncommon orbital cholesterol granuloma of the medial wall approached using a 3D endonasal endoscope

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Abstract

We here described a case of a 57-year-old woman with a sudden onset of mild ptosis and deficit of abduction in her right eye with diplopia. After ophthalmologic evaluation, she performed a CT scan with evidence of a lesion at the level of the orbital medial wall. To determine the nature of this lesion the patient underwent surgery and the surgical excision of the mass was performed through a right side transthmoidal approach using a 3D endoscope. The histological exam revealed a granulomatous reaction and hemorrhagic areas with a rich component of cholesterol clefts and spikes addressing towards a diagnosis of cholesterol granuloma of the orbit.

Keywords: Cholesterol granuloma, orbit, 3D endoscope.

INTRODUCTION

We here describe a case of an orbital cholesterol granuloma in a 57-year-old woman with an uncommon medial wall localization that addressed towards a transnasal approach using a 3D endoscope.

CASE REPORT

A female patient of 57 years old was referred to our Division because of rapidly onset of mild ptosis and deficit of abduction in her right eye with diplopia. After ophthalmologic evaluation, she performed a CT scan with evidence of a lesion at the level of the orbital medial wall and a MRI displaying on T2 a heterogeneous hypo intensity with irregular high signal intensity, hypo intense wall and right orbital compression, and on T1 a heterogeneous hyper intense lesion, located at the right orbital level (Figure 1, a-b).

Since neither clinical nor radiological features addressed towards a neoplastic or infective/inflammatory diagnosis, in order to define the lesion, a surgical

excision of the mass was performed through a right side transthmoidal approach using a 3D endoscope (Visionsense II Ltd, Petach Tikva, Israel).

The lesion, identified at the level of the lamina papyracea, was entirely removed until visualization of the periorbita, and resulted of yellow-brownish fragments encapsulated by bone and adipose tissue with a translucent appearance, for an overall 2,5 cc volume. Postoperative course was uneventful and the patient was discharged 5 days after surgery without ocular manifestation.

The histopatological examination on formalin-fixed paraffin embedded tissue disclosed abundant necrotic material and fibrous tissue with an inflammatory infiltrate. Specifically, lymphocytes and multinucleate giant cells were present in association with a granulomatous reaction and hemorrhagic areas with a rich component of cholesterol clefts and spikes (Figure 2). The diagnosis was cholesterol granuloma of the orbit.

After a 1-year follow-up period the patient has not reported any other diplopia and ptosis episodes, and her

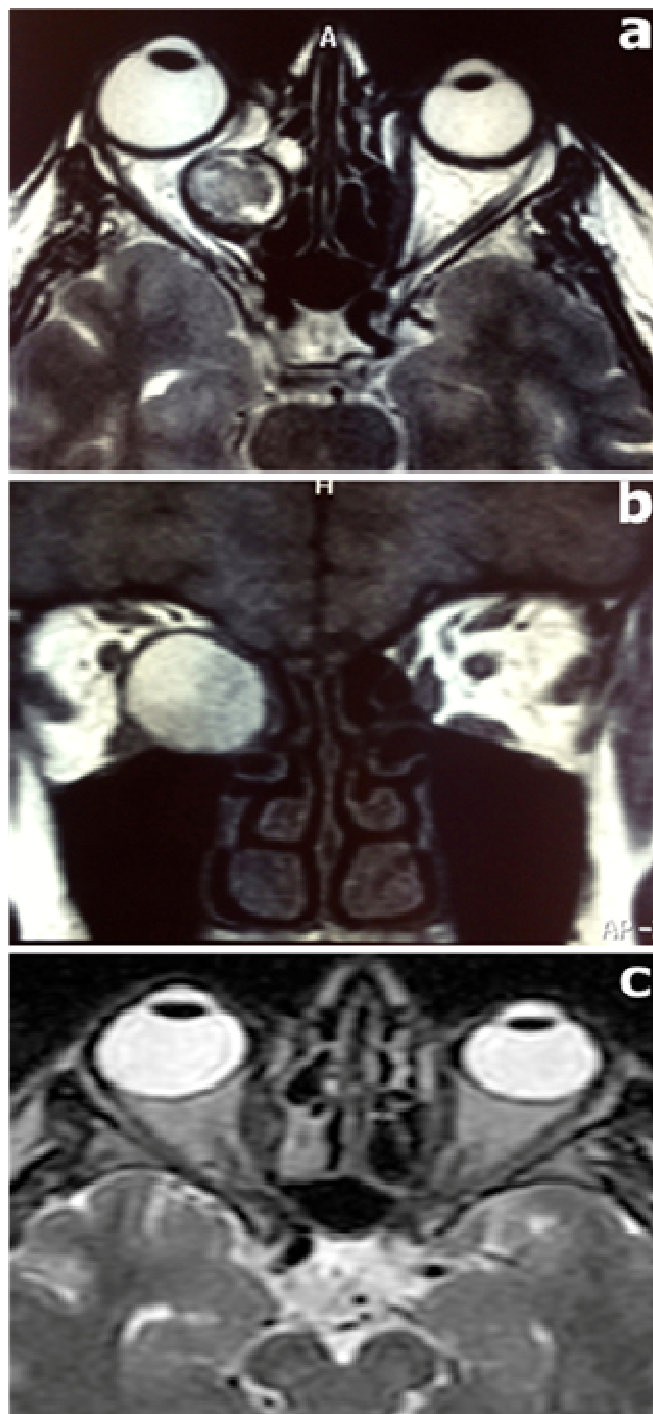


Figure 1, a: MRI of the orbits, axial T2-weighted image showing heterogeneous hypointensity on T2, irregular high signal intensity, hypointense wall with right orbital compression.

Figure 1, b: MRI of facial mass, coronal T1-weighted, visualization of heterogeneous hyperintense lesion on T1, located at right orbital level.

Figure 1, c: MRI of the orbits, axial T2-weighted image showing complete excision of the lesion at the right orbital level.

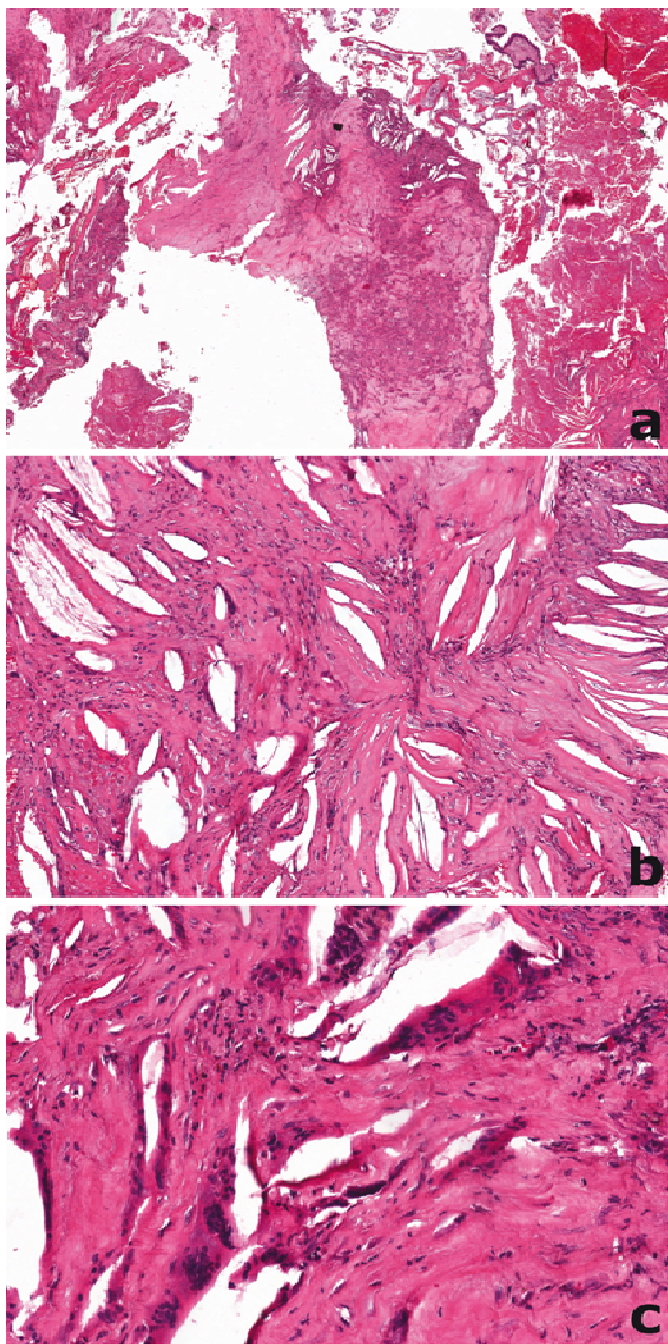


Fig. 2, a: Fibrous connective tissue with characteristic spikes of cholesterol surrounded by multinucleated foreign body giant cells; note the hemosiderin-laden histiocytes immersed in blood field (Haematoxylin-eosin, original magnification $\times 2$).

Fig. 2, b: Inflammatory tissue infiltrated by several cholesterol clefts (Hematoxylin-eosin, original magnification $20\times$).

Fig. 2, c: Granulomatous inflammation with numerous multinucleated giant cells (Hematoxylin-eosin, original magnification $20\times$).

radiological examination MRI (Figure 1,c) showed complete excision of the lesion.

DISCUSSION

Orbital location is very uncommon for cholesterol granuloma (CG), and usually occurs in adults, between the ages of 30 and 60, with a male predilection (McNab and Wright, 1990; Loeffler and Kommerell, 1997). CG are more frequently found in the middle-ear cavity, especially in the mastoid air cell tympanic cavity, which represents the most typical site (Rosca et al., 2006).

At variance to our medial wall site, which addressed towards a trans-nasal endoscopic approach, cholesterol granuloma of the orbital bones usually occurs in the lateral part of the superior orbital ridge within the frontal diploic space (Hill and Moseley, 1992).

Medial wall localization addressed towards a trans-nasal approach using a 3D endoscope, which improves depth perception of field compared to the 2D-endoscope; real-time depth perception becomes of crucial importance, especially in cases of distorted anatomy, since visual perceptual illusion proved to be the primary cause of error in laparoscopic surgery (Way et al., 2003; Roth et al., 2011).

The differential diagnosis of cholesterol granuloma includes mucocele, eosinophilic granuloma, cholesteatoma, multiple types of cysts such as aneurysmal bone cysts, dermoid or epidermoid cysts, cystic ossifying fibroma; they should also be differentiated from lacrimal gland neoplasms.

Typical clinical and radiologic features help differentiate most of these conditions from orbital cholesterol granulomas before surgery (Hill and Moseley, 1992).

Orbital cholesterol granulomas have characteristic radiologic imaging findings: CT usually shows an isodense to hypodense osteolytic lesion, or, more rarely, a cystic mass lesion eroding the bone; MRI typically shows a no-contrast enhancing lesion with high signal intensity on both T1- and T2-weighted images, with a high signal intensity on fluid-attenuated inversion recovery ('FLAIR') MRI and low signal intensity on diffusion-weighted MRI (Hill and Moseley, 1992).

Cholesterol granuloma seems to be caused by a foreign body reaction around cholesterol crystals: in 1838, Muller introduced the term "cholesteatoma" for any lesion with cholesterol crystals and spikes (Fukuta and Jackson, 1990; Arat et al., 2003).

Parke et al suggested to characterize between "epidermoid or true cholesteatoma" referring to lesion containing epithelial elements and "cholesterol granulomas" in case of non-epithelium containing pseudotumours (Parke et al., 1982). Both lesions present cholesterol clefts and foreign body multinucleate giant

cells along with blood degradation products (Arat et al., 2003).

Differential diagnosis between these two entities is relevant due to a different recurrence risk: approximately 28% of orbital cholesteatoma recur with the possibility of malignancy found at the time of re-exploration. Therefore, the appropriate surgical technique in both cases is complete resection, since the differential diagnosis is exclusively histological and cannot be performed prior to surgery. The effectiveness of endonasal endoscopic surgical procedures has increased considerably with the introduction of three-dimensional vision. Working in a three dimensional environment allows to be more selective and precise during all surgical steps. Surgical gesture is increased also during the nasal approach preserving the olfactory mucosa in order to reduce the rate of post-operative anosmia.

Nowadays the pathogenesis of CG remains unclear: past or remote trauma could be a provoking cause with the subsequent bleeding. The presence of cholesterol within the lesion is believed to be consequence of haemorrhage, as a result of the breakdown of blood components and this is followed by foreign body granuloma formation (Arat et al., 2003). Other authors, instead, support the concept of airway obstruction in the well pneumatized cells of temporal bone and paranasal sinus. The coagulated blood is retained in the frontal sinus, causing obstruction of air drainage: the union of blood products and the deposition of cholesterol crystals finally causes cholesterol granuloma (Maniglia and Villa, 1977; Rosca et al., 2006). Orbital subperiosteal space has a unique anatomy and susceptibility to various pathological process, since this virtual space allows the slow reabsorption of the hematoma and the formation of degradation products of hemoglobin resulting in cholesterol granuloma.

In conclusion, orbitofrontal cholesterol granulomas, though rare in this site, should always be suspected when cholesterol clefts, foreign body multinucleate giant cells with blood degradation products are present in absence of an associated epithelial component.

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