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Original Article

Clinical, radiological and histopathological characteristics of surgically removed orbital hematic cysts: A case series

Hind M. Alkatan [a,b,*]; Mohammed D. Alotaibi [a]; Azza M.Y. Maktabi [c]; Hamad Aljaedi [b]; Sahar M. Elkhamary [d,e]; Yasser Al-Faky [a]; Adel H. Alsuhaibani [a]

Abstract

Background: Hematic cyst is a rare orbital condition that has a wide range of clinical presentation and is characterized pathologically by lack of endothelial lining.
Purpose: To correlate clinical and radiological features of hematic cysts, to tissue diagnosis, and investigate the possible etiology behind this condition, its relation to trauma and other interesting histopathological findings.
Methods: Retrospective case series at King Khaled Eye Specialist Hospital (KKESH) and King Abdulaziz University Hospital (KAUH) of all orbital lesions with tissue findings supporting the clinical and/or radiological diagnosis of hematic cyst.
Results: A series of 13 cases was studied, 8 males and 5 females. Age ranged from 2 to 84 years with a median of 54. Most cases presented with proptosis (76.9%) and limitation of eye movements (69.2%). History of trauma was confirmed in only 2/13. The clinical diagnosis of hematic cyst was made prior to surgery in 38.4%. Magnetic Resonance Imaging (MRI) confirmed the presence of blood in the orbit in 7/7. Surgical intervention was the mainstay of treatment. Histopathologically, these lesions demonstrated variable constituents including blood break-down products (hemosiderin), macrophages, mononuclear inflammatory cells, hemorrhage, absent endothelial lining, reactive fibrosis and capsule-like formation. Cholesterol clefts with typical granulomas and multinucleated giant cells were present in 2 cases. A clue to an underlying vascular lesion was found histopathologically in 30.8%. None of the patients developed recurrence or long-term complications with an average follow up period of 1 year.
Conclusion: Hematic cyst is a challenging clinical diagnosis that can be aided by radiological examination and histopathological confirmation. Trauma does not seem to play a major role while presence of a pre-existing vascular lesion with spontaneous hemorrhage may be an etiologic factor. Associated cholesterol granuloma is an interesting controversial finding. Surgical intervention is curative with possible persisting motility disturbance and/or the eye deviation and worse prognosis in post-traumatic cases.

Keywords: Orbital, Hematic cyst, Hemorrhage, Vascular, Granuloma, Hemosiderin

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Introduction

Hematic cyst is a term used to describe a blood-derived lesion in the orbit.[1] The characteristic features to define hematic cysts compared to other blood-derived lesions is actually the lack of endothelial lining and the fact that they are encapsulated by a layer of fibro-collagenous material.[2] Hematic cyst is considered a rare condition and usually

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[a] Department of Ophthalmology, King Saud University, Riyadh, Saudi Arabia
[b] Department of Pathology, King Saud University-Medical City, Riyadh, Saudi Arabia
[c] Pathology and Laboratory Medicine Department, King Khaled Eye Specialist Hospital, Riyadh, Saudi Arabia
[d] Diagnostic Imaging Department, King Khalid Eye Specialist Hospital, Riyadh, Saudi Arabia
[e] Department of Diagnostic Radiology, Mansoura Faculty of Medicine, Mansoura, Egypt

* Corresponding author at: Departments of Ophthalmology and Pathology, College of Medicine, King Saud University, Riyadh, Saudi Arabia.
e-mail addresses: hkatan@ksu.edu.sa, hindkatan@yahoo.com (H.M. Alkatan).

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presents with symptoms due to its anatomical effect in the orbital cavity that includes proptosis, diplopia, visual disturbance and strabismus.[3]

Orbital imaging has played a major rule in the diagnosis of hematic cyst. Computerized tomography (CT) imaging typically shows a well-demarcated homogeneous mass but can show heterogeneity with layering of blood products in some cases. MRI is the imaging modality for the evaluation of the cyst nature because it is useful in confirming the presence of blood in the lesion as well as in assessing the stage of hemorrhage according to the signal intensity variation depending on the age of the hemorrhage.[4]

The aim of this retrospective study is to establish a correlation between the clinical, radiological, and pathological findings that can aid in a better understanding of these lesion, investigate the possible etiology and provide better approach to the management of such cases.

Methods

This is a retrospective case series including all patients diagnosed with hematic cyst by histopathological examination after undergoing surgical removal at 2 Eye centers in Saudi Arabia: King Khaled Eye Specialist Hospital (KKESH) and King Abdulaziz University Hospital (KAUH). Thirteen cases where included from the histopathology data base of the 2 institutions over the period 2000–2017. The demographic data (age, sex and nationality) and basic clinical information was collected by chart review. The pre-operative radiological studies were reviewed by an experienced neuro-radiologist. Data was completed by histopathological review of the histologic sections and the reports to confirm the clinical and/or the radiological diagnosis of hematic cyst, identify the cases with the specific findings of a cholesterol granuloma and to observe any clue suggestive of an underlying vascular lesion or anomaly.

Results

We had a total of 13 cases with histopathological findings that are consistent with the clinical and/or radiological impression of an orbital hematic cyst. 8 cases were males and 5 were females. The age ranged from 2 to 84 years with a mean of 44 years and median of 54 years. The right orbit was involved in 9/13.

History of trauma was confirmed in 2 cases only (15.3%). One of these was a 9-year old boy with history of undefined trauma few weeks prior to his presentation with proptosis and blurred vision on the left side (Case 4). He had an extensive hematic cyst extending to the left orbital apex with complete loss of vision in the affected eye, relative afferent pupillary defect (RAPD), and frozen globe. The other patient (Case 10) was a 70-year-old healthy lady presenting with stable left eye protrusion 4 months following a history of fall and facial trauma. She had counting finger vision in that eye, RAPD and limitation of supraduction and abduction. History of surgery (without trauma) was documented in one 25-year old patient who was 6 months post-secondary orbital implant (Case 7). One case had worsening of symptoms following therapeutic Botox injection for squint management (Case 9). No clinical history of a pre-existing orbital vascular lesion was found in any of the cases. Summary of the cases is provided in Table 1.

Proptosis was the commonest presenting symptom in 10/13 (76.9%) followed by limitation of eye movements in 9 cases (69.2%).Other less common unusual presentations included decreased vision in the affected eye (4/13), upper lid swelling (3/13), socket-related bleeding after enucleation and squint in one patient each (case 7 and case 9 respectively). Interestingly, the clinical diagnosis of hematic cyst was made prior to the surgery in 5 cases (38.4%), in whom the impression was even questionable.

Radiological imaging was performed in a total of 10 patients as summarized in Table 1 and revealed an intraconal lesion in 6/10. The 7 cases who underwent MRI had confirmation of the presence of blood in the orbit, thus the impression of hematic cyst was raised. CT scan was performed in 3 cases, 2 of which were extraconal lesions. The clinico-radiological findings of 3 cases are demonstrated in Fig. 1–3 in addition to Fig. 4A through D.

Surgical intervention was the mainstay of treatment with a variable surgical orbitotomy approach depending on several factors most importantly the location of the lesion. Histopathologically all lesions demonstrated collection of blood (often thrombosed) with absent endothelial lining, variable proliferating reactive fibroblasts, and surrounding formation of a fibrous capsule (Fig. 4E). Other constituents included chronic inflammatory cells in 4/13 cases and blood break-down products (hemosiderin) in 4/13(Fig. 4F). We had 2 unique cases of cholesterol granulomas showing typical cholesterol clefts, hemosiderin-laden macrophages, and multinucleated giant cells (Cases 11 and 12, one of which is shown in Fig. 5A through D). A histopathological clue to an underlying vascular lesion (lymphatic/venous malformation in 3 and lymphangioma in 1) was found histopathologically in 30.8% of the cases. None had confirmed history or confirmed clinical diagnosis of this pre-existing vascular lesion prior to the recent presentation. None of the patients developed recurrence or long-term complications with a follow up period up to 1 year except for motility-related sequelae.

Discussion

We have aimed to study hematic cysts in the orbit since they are poorly understood with vague etiology. Shiparo and colleagues drew a connection of similarity between subdural hematoma and orbital hematic cysts.[5] Bleeding in the orbit can be generally traumatic or non-traumatic. Spontaneous or non-traumatic orbital hemorrhage can be associated with other predisposing factors such as orbital vascular malformations, bleeding disorder, or sudden increase in venous pressure. This underlying presumed vascular malformation was found in approximately one third of our cases (30.8%) while history of trauma was confirmed in 2 patients only. Many theories have speculated other causes of bleeding in the absence of vascular malformations. Non-traumatic orbital hemorrhage was classified by McNabb into diffuse intraorbital, localized intraorbital (hematic cyst), subperiosteal, extraocular muscle (EOM)-related and orbital floor implants-related.[6] According to the previously mentioned classification, we had 11/13 non-traumatic orbital hemorrhage cases, out of which 10 were considered localized intraorbital hematic cyst and one was sub-periosteal. Also,