Hematic Pseudocyst Masquerading as Orbital Cellulitis and Sinusitis

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Abstract
Hematic pseudocysts are fibrous, nonepithelial lined capsules containing blood byproducts that typically present remotely following orbital fracture hardware implantation. Trauma, implant migration, and tissue erosion are believed to cause hemorrhage to pool within the capsular space. Risk factors include inadequate posterior fracture reduction and use of nonporous material which prevents fibrovascular ingrowth and stabilization. Mass effect from these lesions may cause patients to present with pain, lid swelling, hyperglobus, proptosis, lid retraction, motility restriction, or blurry vision. Pseudocysts associated with fracture hardware have been misdiagnosed as tumors or in one prior case as an infection. Herein we report a unique case of hematic pseudocyst masquerading as orbital cellulitis with maxillary sinusitis. A 59-year-old man presented with periorbital pain, hyperglobus, proptosis, and ptosis 2 years after repair of an orbital floor fracture. CT demonstrated a soft tissue collection adjacent to an implant as well as maxillary sinus opacification. He did not improve with antibiotics, at which point surgery revealed a pseudocyst and its contents were removed. This report describes a unique presentation of orbital pseudocyst and summarizes the literature on this entity.

Introduction
Hematic pseudocysts are fibrous, nonepithelial lined capsules containing blood byproducts [1]. They have been referred to as “hematic cysts” or “chocolate cysts,” though they are not true cysts given their lack of an epithelial lining. The term “cholesterol granuloma” has also used interchangeably, though it has been applied more in nontraumatic and nonorbital cases [2].
More than 90% of orbital pseudocysts occur in the superotemporal or superior orbit, either spontaneously or in response to trauma. There have been no reports of superonasal or medial orbital pseudocysts, but rare reports of intracanal pseudocysts exist. Orbital floor cysts typically occur around implants used to repair fractures and may occur as late as 25 years following implantation [2–4].

The precise mechanism of capsule formation and fluid accumulation remains uncertain. A capsule may form around an implant, creating a space for blood and fluid to accumulate if trauma and implant migration cause erosion of the periosteum, infraorbital vessels, and lymphatic outflow. It has also been postulated that incomplete fluid resorption causes chronic granulomatous inflammation, which in turn induces formation of the capsule [5]. Other studies indicate that pseudocysts form via a similar mechanism as chronic subdural hematomas, where increased fibrinolysis from tissue plasminogen activator in the neomembranous lining causes chronic low grade flow [1, 6]. Risk factors for pseudocyst formation include inadequate posterior fracture reduction, which causes chronic irritation of the periosteum and capsule, and use of nonporous material, which prevents fibrovascular ingrowth and stabilization [5, 7].

Mass effect from these lesions causes patients to present with pain, lid swelling, hyperglobus, proptosis, lid retraction, motility restriction, and blurry vision. Imaging typically reveals a well-defined, homogenous mass surrounding the implant and prior fracture [8]. Surgical decompression and excision of the capsule, with or without removal or replacement of loose hardware, can alleviate globe displacement and its resultant symptoms [5].

Herein, we report a unique case of hematic pseudocyst masquerading as orbital cellulitis with maxillary sinusitis. Written, informed consent was obtained from the patient, and all patient information was handled in compliance with HIPAA guidelines.

**Case Report/Case Presentation**

A 59-year-old male presented to an outside Emergency Room 2 years after orbital floor fracture repair reporting 2 days of right-sided periorbital pain and swelling. He denied recent trauma, discharge, fever, rash, or vision changes. He reported chronic nasal allergies but no sinus pain, congestion, or sore throat. CT showed a soft tissue collection between the inferior rectus and floor fracture fragment with peripheral enhancement and central fluid density as shown in Figure 1. Maxillary sinus inflammation was also noted. The patient was diagnosed with orbital cellulitis, started on intravenous antibiotics, and transferred to our institution.

![Fig. 1. Coronal (a) and sagittal (b) CT scan demonstrating fluid collection in the inferior orbit (asterisks) and maxillary sinus (arrow).](image-url)
Upon our examination, visual acuity was 20/20, and intraocular pressure, pupils, and motility were normal. There was mild periorbital edema, hyperglobus, proptosis, and ptosis as demonstrated in Figure 2. The rest of the eye exam was normal. His lack of fever or infectious symptoms raised suspicion for a noninfectious process. He was admitted and continued on intravenous antibiotics for 1 day without improvement, at which point a decision was made to drain the fluid collection and explant the associated hardware.

Transconjunctival orbitotomy was performed, revealing two stacked silicone/plastic implants with titanium screw fixation loosely overlying the orbital floor fracture. The implants were surrounded by a blood byproduct-filled capsule without purulence. Incomplete posterior fracture reduction was noted. The implant, screw, and blood were removed from the orbit. The globes were then found to be symmetric without enophthalmos, so no additional implant was placed and the incisions were closed. The patient was discharged on an oral and topical antibiotic and made a fast and complete recovery.

Discussion/Conclusion

Twenty-seven prior cases of hematic pseudocysts after hardware implantation for an orbital fracture have been reported and were available for full-text review. Porous materials such as porous polyethylene are thought to reduce the risk of pseudocyst formation by allowing fibrovascular ingrowth which stabilizes the implant [5]. Retrospective studies by Peng et al. [7] and Romano et al. [8] reported 1/269 and 0/140 incidence of pseudocyst or hematoma with porous implants. However, pseudocysts are known to occur many years after surgery and only 22% of the patients in Peng et al.'s [7] study followed up for more than 12 weeks. Six recent cases have demonstrated pseudocyst formation with porous materials [5, 9–11]. Our patient's stacked, nonporous implants were found to be loosely screwed to bone and relatively mobile, likely contributing to hemorrhage and capsule formation.

Given their rarity, pseudocysts may be misdiagnosed as other mass lesions. Chao et al. [12] described an inferior pseudocyst initially thought to be a choroidal tumor given apparent globe indentation on fundoscopy. A similar report described two young men diagnosed with malignant tumors prior to surgical exploration which revealed pseudocysts [13]. Gupta et al. [4] published the one prior case of misdiagnosis as an infection. A 65-year-old man developed recurrent episodes of vertical diplopia over the course of 1 year which were initially attributed to transient ischemic attacks. After eventually developing symptoms concerning for orbital cellulitis, surgery was undertaken and demonstrated a large pseudocyst surrounding a 25-year-old implant for a fracture. Pseudocyst expansion into the maxillary sinus has been described in one prior case but not in association with orbital hardware [14].
To our knowledge, this is the first reported case of a hardware-associated orbital pseudocyst masquerading as orbital cellulitis and sinusitis. Inadequate posterior fracture reduction likely allowed communication of the cyst with the maxillary sinus, but also contributed to pseudocyst formation. Orbital surgeons should be aware of the increased risk with nonporous implants, the potential for pseudocyst formation decades after hardware placement, and the possibility of misdiagnosis as infectious or neoplastic entities.

Statement of Ethics

This research publication complies with the Declaration of Helsinki. Ethics Committee and IRB approval were not required in accordance with the policies of our institution (https://www.uchealth.org/wp-content/uploads/2019/12/UCHealth-IRB-SOP-Nov-2019.pdf). Written informed consent to publish case history and images was obtained from the patient.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Ryan Larochelle, Alexandra Levitt, and Sophie Liao each participated in the care of this patient and the drafting and editing of this manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article.

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