

Brief Report

Rescue technique for complete removal of an accidentally ruptured orbital dumbbell deep dermoid cyst: A case report

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ABSTRACT

Purpose: To report a rescue technique for complete removal of an accidentally ruptured orbital dumbbell deep dermoid cyst.

Observations: A 33-year-old female presented with left proptosis with retrobulbar discomfort for 3 months. Computed tomography images showed an orbital dumbbell deep dermoid cyst. A lateral orbitotomy was performed under general anesthesia. The cyst was ruptured during osteotomy of the lateral orbital rim. The cyst was opened vertically from the ruptured site using a Stephen's tenotomy scissors to visually confirm the internal wall of the cyst and to keep the epithelial lining intact while separating the external wall of the cyst from the bone. There was a small defect of the epithelial lining at the inferoposterior margin of the cyst. Granulomatous inflammation of the lacrimal gland was found adjacent to the defect site. The cyst was completely removed and the lacrimal inflammation subsided after the operation.

Conclusions and importance: Opening of the cyst is a useful technique to remove the entire epithelial lining of an accidentally ruptured dumbbell dermoid cyst.

1. Introduction

Deep orbital dermoid cysts are choristomas with a predilection for the superotemporal and superonasal osteosutures.¹ These cysts are usually asymptomatic until adulthood, and present with a gradual painless proptosis or an ocular motility problem. They sometimes rupture spontaneously, which causes inflammation of the eyelids and orbits.² A complete removal is mandatory to avoid postoperative recurrence and inflammation.³

The dumbbell dermoid cyst is a subtype of the deep dermoid cyst. Several techniques have been reported to avoid unintended intraoperative rupture of this entity, but these techniques are often difficult.³ In addition, it has been rarely mentioned how to facilitate complete removal after an intraoperative rupture.

We, therefore, present a case showing a rescue technique to facilitate complete removal of an accidentally ruptured dermoid cyst by initially opening the cyst.

2. Case report

A 33-year-old woman presented with left proptosis with retrobulbar discomfort for 3 months (Fig. 1). Examination revealed a fixed, non-

tender, firm palpable mass in the superolateral aspect of the left orbit rim. The left best-corrected visual acuity was 1.2. There was an inferior displacement of the left globe with no restriction of ocular motility. Hertel exophthalmometry measurements were 15.5 mm on the right side and 22 mm on the left side, with a base of 108 mm. Computed tomography (CT) showed a well-defined, thin walled, 2-component cyst in the intraconal and extraconal spaces in the superolateral aspect of the left orbit, measuring 30 × 26 × 32 mm, with an erosion of the greater wing of the sphenoid and the frontal bone (Fig. 2). The cyst extended from the orbit to the left temporal fossa through the defect in the lateral wall of the orbit. The preoperative diagnosis was dumbbell dermoid cyst of the left orbit and temporal fossa.

Lateral orbitotomy was performed under general anesthesia to excise the intra- and extraorbital components in toto. The periosteum of the lateral orbital margin was exposed, and then, was incised and reflected to expose the lateral orbital rim. The lateral orbital rim was temporally removed with a pneumatic oscillating saw, followed by the use of a hammer and chisel. When the rim was fractured using a rongeur, the cyst was ruptured and some oily materials with hairs spilled out, which were suctioned and irrigated. The cyst was opened vertically from the ruptured site using a Stephen's tenotomy scissors to visually confirm the internal wall of epithelial lining, and the external wall of

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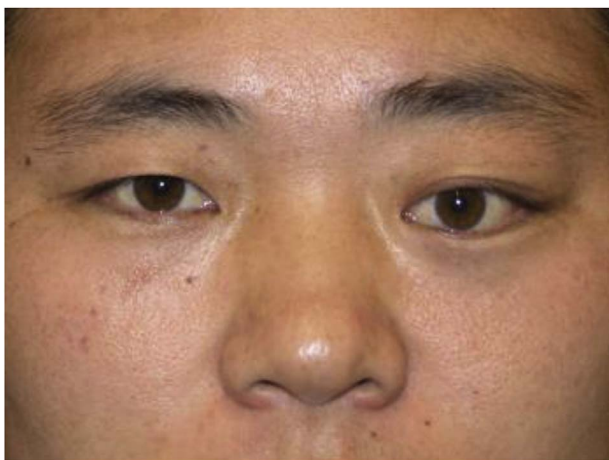


Fig. 1. A preoperative patient's face photo showing left proptosis and inferior globe displacement.

the cyst was separated from the bone with keeping the epithelial lining intact (Fig. 3A). There was a small defect of the epithelial lining at the inferoposterior margin of the cyst. After removing the epithelial lining completely (Fig. 3B and C), an unusual hard tissue was identified in the lacrimal fossa, which was also removed. The bone defect was covered using a 0.3 mm-thick poly-L-lactic acid/hydroxyapatite sheet and the temporally removed lateral orbital rim was repositioned and fixed using poly-L-lactic acid/hydroxyapatite plates (both, Super Fixsorb Mx[®]; TELJIN MEDICAL TECHNOLOGIES CO., LTD; Osaka; Japan).

Histopathologic examination of the excised lesion confirmed dermoid cyst with adnexal structures such as sebaceous glands and hair shafts (Fig. 4A). The inside of the cyst was lined with stratified squamous epithelium. The outside surface was thickened with inflammatory tissue (Fig. 4B). The hard tissue in the lacrimal fossa was a part of the lacrimal gland with granulomatous inflammation.

CT images taken in the postoperative following day showed no residual cyst (Fig. 5A and B). At postoperative 2-month follow-up, the inferior displacement of the globe and proptosis improved but still somewhat remained (Fig. 5C). The lacrimal inflammation had subsided.

At postoperative 8-month follow-up, Hertel exophthalmometry measurement decreased to 18 mm on the left side. The extraocular motility was normal.

3. Discussion

We presented a rescue technique of complete removal of an accidentally ruptured orbital dumbbell deep dermoid cyst by initially opening the cyst. Removal of the total epithelial lining avoids postoperative recurrence and inflammation.³ The vertical incision of the surgically ruptured site provided a larger view of the lining, which facilitated complete removal of the epithelium.

Sclerotherapy has been described as a minimal invasive technique for periorbital dermoid ablation.⁴ This technique eliminates epithelial cells within the cyst and leaves a collapsed cyst wall within the tissue.⁴ However, in some of cases with solid contents of a dermoid cyst, drainage of cyst contents before sclerotherapy is difficult, and the solid contents may prevent successful sclerotherapy.⁵

En block removal of the orbital dumbbell dermoid cysts is often difficult as they are tightly adhered to the bones at the frontozygomatic suture.^{1,2,6} Therefore, an accidental cyst rupture occasionally occurs, which also happened in the present case. Several surgical techniques have been reported to avoid unintended surgical rupture. Prior removal of a small bone fragment next to the bony defect is effective for preventing rupture.⁶ The prolapsed extraorbital component can be reduced separately, followed by the removal of intraorbital component.^{1,7} Decompression by needle aspiration facilitates the exposure of the cyst.⁸ Methylene blue injection delineates the extent of the cyst.⁴ These techniques can be combined with opening the cyst.

4. Conclusions

Initial opening of the cyst was a useful technique to remove the entire epithelial lining of an accidentally ruptured dumbbell dermoid cyst.

Patient consent

Institutional review board approval and written patient consent to

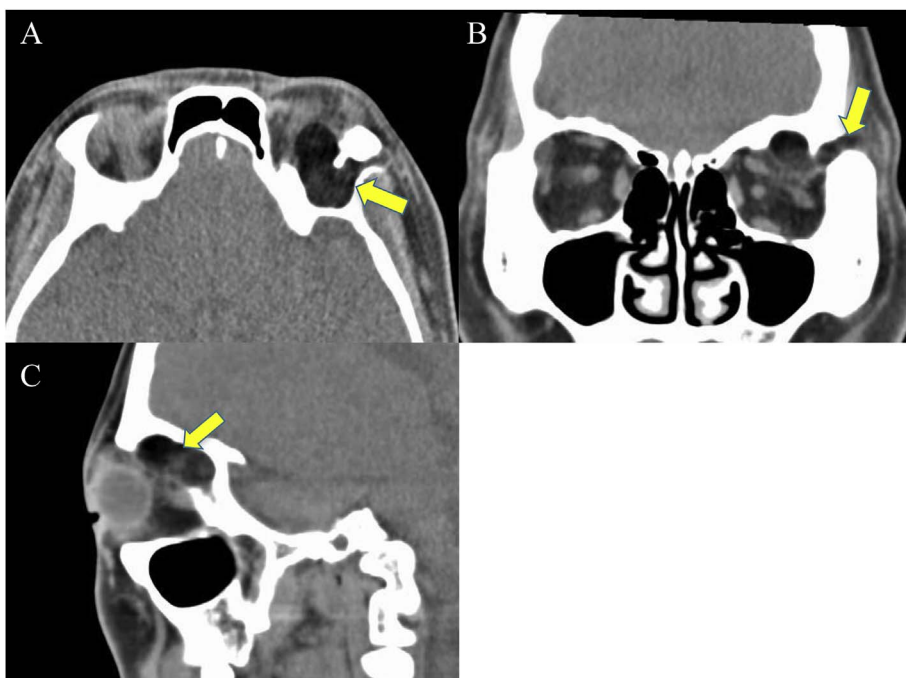


Fig. 2. Preoperative (A) axial, (B) coronal, and (C) sagittal sections of computed tomographic images. A well-defined, thin walled, 2-component cyst in the extraconal space in the superolateral aspect of the left orbit near the lacrimal fossa (arrows).

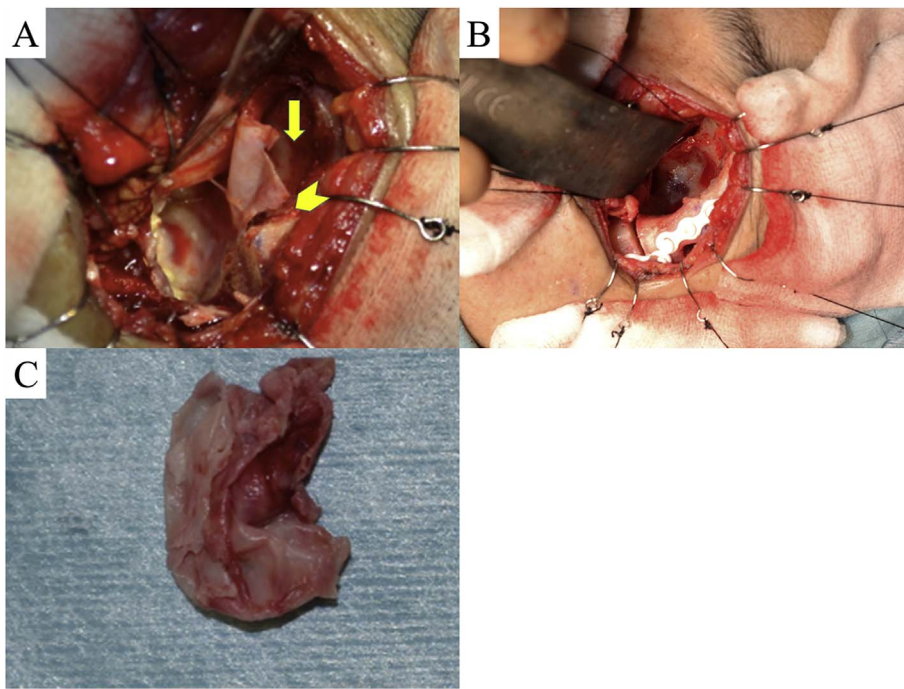


Fig. 3. Intraoperative photographs. (A) The epithelial lining on the internal surface of the cyst is exposed (arrow). The cyst is cut vertically for larger view of the lining (arrowhead). (B) After completion of the cyst removal. (C) Macroscopic appearance of the removed cyst.

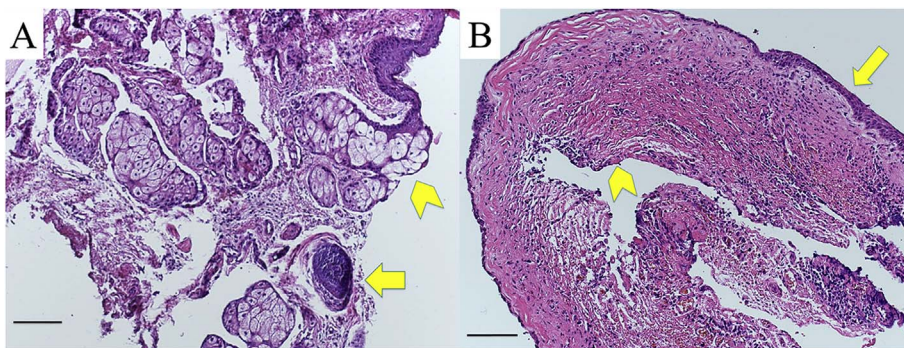


Fig. 4. Pathologic findings. (A) Hair shaft (arrow), sebaceous gland (arrowhead), and stratified squamous lining (hematoxylin and eosin x 100), (B) Stratified squamous epithelial lining on the internal surface of the cyst (arrow) with thickened outer layer of foreign body giant cells (arrowhead) (hematoxylin and eosin x 100) (bar length = 100 μ m).

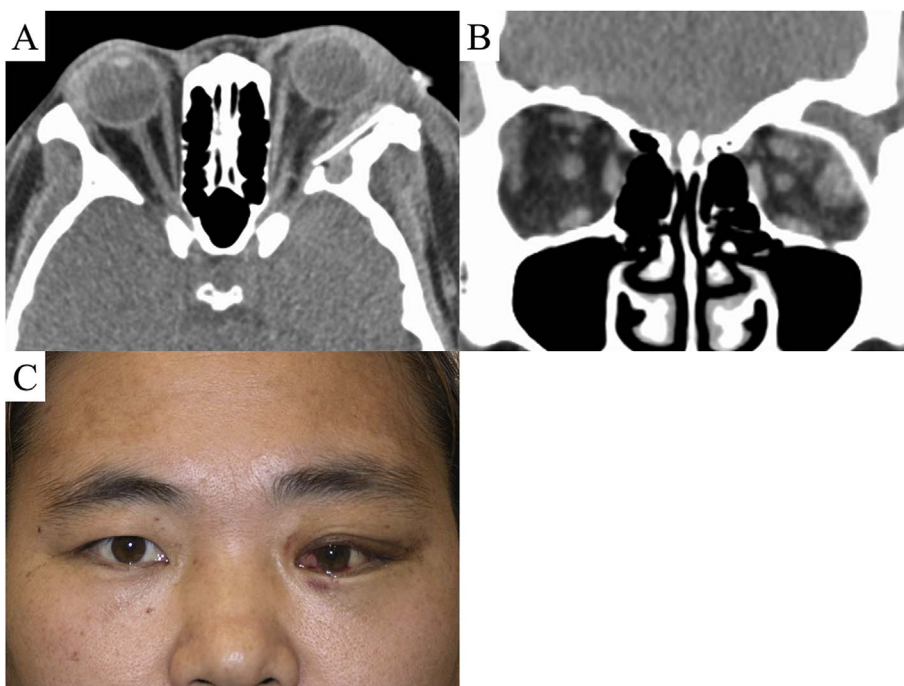


Fig. 5. Postoperative findings. Postoperative (A) axial and (B) coronal sections of computed tomographic images. The cyst is completely removed. The bone defect is covered using an absorbable sheet. (C) A patient's face photo taken at postoperative 2 months.

publish case details such as photographs in this case report have been obtained.

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Conflicts of interest

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Authorship

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Appendix A. Supplementary data

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.ajoc.2018.01.044>.

References

1. Shields JA, Kaden IH, Eagle Jr RC, Shields CL. Orbital dermoid cysts: clinicopathologic correlations, classification, and management. The 1997 Josephine E. Schueler Lecture. *Ophthalmic Plast Reconstr Surg*. 1997;13(4):265–276.
2. Siah WF, Al-Muhaylib AA, Rajak S, et al. Clinical outcomes of ruptured periorbital and orbital dermoid cysts. *Ophthalmic Plast Reconstr Surg*. 2017;33(4):264–267.
3. Sherman RP, Rootman J, Lapointe JS. Orbital dermoids: clinical presentation and management. *Br J Ophthalmol*. 1984;68(9):642–652.
4. Naik MN, Batra J, Nair AG, et al. Foam sclerotherapy for periorbital dermoid cysts. *Ophthalmic Plast Reconstr Surg*. 2014;30(3):267–270.
5. Gupta VP, Gupta P, Gupta R. Re: “foam sclerotherapy for periorbital dermoid cysts”. *Ophthalmic Plast Reconstr Surg*. 2014;30(6):530.
6. Yuen HK, Chong YH, Chan SK, et al. Modified lateral orbitotomy for intact removal of orbital dumbbell dermoid cyst. *Ophthalmic Plast Reconstr Surg*. 2004;20(4):327–329.
7. Samuelson TW, Margo CE, Levy MH, Pusateri TJ. Zygomaticofrontal suture defect associated with orbital dermoid cyst. *Surv Ophthalmol*. 1988;33(2):127–130.
8. Meyer DR, Lessner AM, Yeatts RP, Linberg JV. Primary temporal fossa dermoid cysts. Characterization and surgical management. *Ophthalmology*. 1999;106(2):342–349.