

Surgical Procedure and Perioperative Management of a Giant Orbital Echinococcus Cyst during and after Pregnancy

Chirurgisches und perioperatives Management einer sehr großen orbitalen Echinococcus-Zyste während und nach einer Schwangerschaft



Introduction

Orbital cystic echinococcosis (CE) is a rare entity and most common in poor countries. Surgical management has been published in a few case reports [1–6]. About 0.8–1 percent of all CE cases show an orbital involvement [4] and present typical radiological findings [6].

Interdisciplinary workup (radiology, infectious diseases) is usually performed before surgery and antiparasitic therapy is started before surgery in consideration of intraoperative spilling and a possible anaphylactic reaction, which is caused by the spilling of fluid from the hydatid cyst [7]. The prevalence of anaphylactic shock during surgery varies among studies and might be around 2% [8].

History and Signs

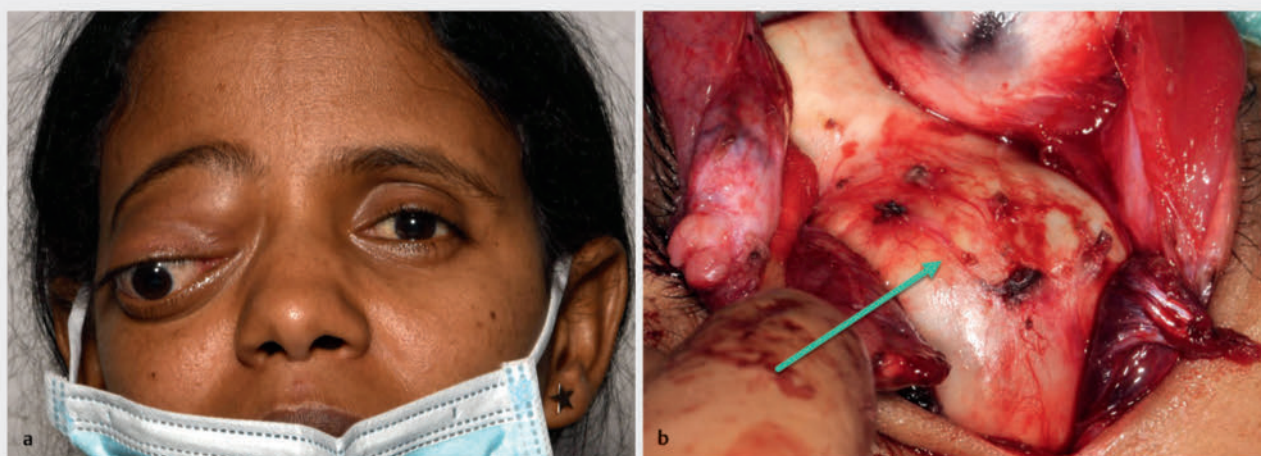
We present, to the best of our knowledge, the first case of a pregnant woman and the surgical and interdisciplinary consequences for management of a highly suspicious orbital CE over the whole course of early and late pregnancy, natural birth, and breastfeeding.

A 29-year-old woman, a refugee from Eritrea who just recently immigrated to Switzerland, was sent to our hospital with a giant orbital cystic lesion (► Fig. 1 a). She had been suffering from this lesion for around 2 years. She was underweight (body mass index 16.6), with no other known diseases, and mother of a 4-year-old child.

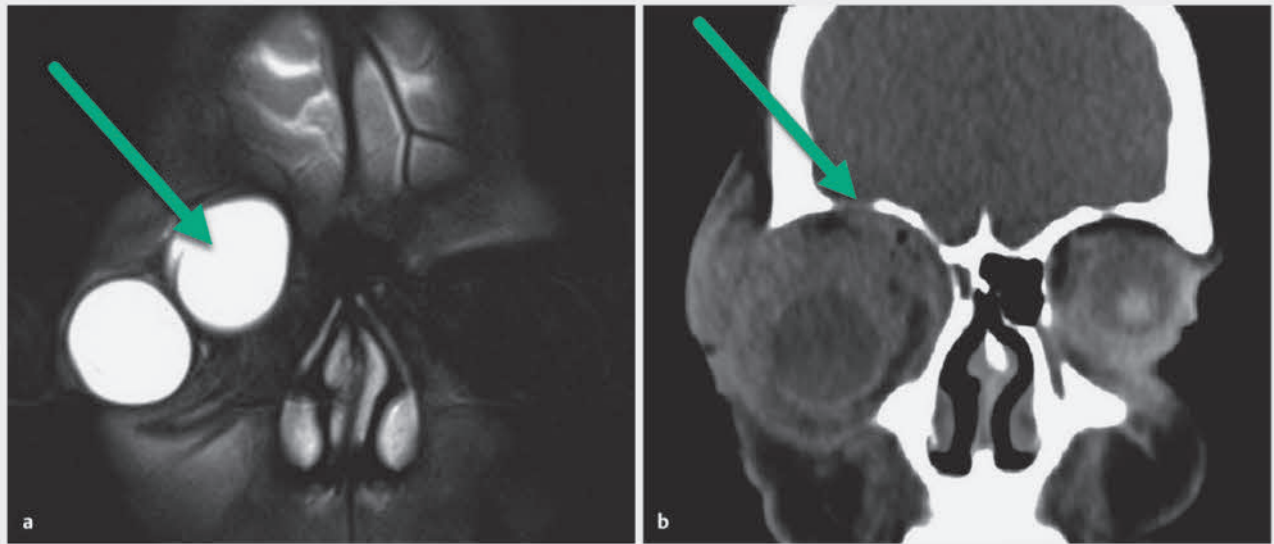
The right eye showed a gigantic protrusion with inferolateral globe displacement. Lateral, upper, and inferior eye movements were possible, reduced medially. Visual

acuity was hand movements, and a lagophthalmus of 3 mm, without corneal staining, due to positive Bell's phenomenon. The optic nerve showed advanced atrophy, no disc swelling, and matching to a longer lasting process. Because the lesion was rather displacing the globe and not infiltrating the eye and because of the longer history, a more benign process was suspected. The left eye showed no pathological finding, full vision, and a normal visual field examination.

Imaging magnetic resonance tomography (MRT) revealed suspicion of a giant orbital CE (► Fig. 2 a). Further general checkup showed present IgG for hepatitis A and B, but no cysts within the liver were found. Just before the planned surgery, a repeated checkup revealed a pregnancy in the 12th week. This initiated a discussion of the best surgical and anthelmintic management for the mother and the fetus, since the parents did not want to have



► Fig. 1 a Preoperative clinical aspect. Right eye shows protrusion and temporal inferior displacement. b Intraoperative aspect seen from above. Green arrow points out the huge yellowish cyst with a thick rubber-like membran.



► **Fig. 2** a MRI T2-weighted preoperative image (coronal plane) shows a hyperintense cyst medially in the right orbit and temporal displacement of the eye. b CT scan (coronal plane) just before surgery. Green arrow points to erosion of the orbital roof.

any risks for the unborn child and the patient herself was highly underweight due to suffering from pregnancy emesis. In the interdisciplinary discussion, surgery under antiparasitic medication was strongly recommended by the infectiologists, fearing the cyst would rupture, with cerebral spreading of the infection. The neonatal unit recommended waiting until the 34th week of pregnancy because after the 34th week, a newborn is thought to have a higher chance of survival due to lung maturation.

Therapy and Outcome

As the optic nerve was already atrophic and the situation for the patient well manageable, we decided, together with the patient, the risk of surgery was too high to take before birth for mother and the unborn child due to general risks of such a long surgery, risk of anaphylactic shock by spilling, risk of cerebral bleeding, as the bony orbital roof was atrophic, and medication-related side effects (e.g., diarrhea, vomiting).

We performed monthly follow-up examinations in special regard to the healthy contralateral left eye. Visual acuity on the right eye stood stable at hand movements.

Over the course of 7 months, the lesion grew slowly and local dryness was well controlled with moisturizing ointment.

A healthy baby was naturally born complication free in an external hospital and no rupture of the cyst occurred. Since the patient was still very underweight, as during her first pregnancy, felt rather weak after birth, and also wanted to breastfeed, surgery was postponed by the patient until she stopped breastfeeding and felt stronger and ready for the surgery, which was 4 months later. Surgery was then planned in close cooperation with the infectious diseases department. Oral anthelmintic medication was initiated with albendazole (400 mg 24 hours before surgery) and praziquantel (3000 mg 24 hours before surgery). Preoperative imaging showed a direct attachment of the cyst to the dura in the superior orbit due to the atrophy of the bony orbital roof (► **Fig. 2 b**).

The surgical approach was started through a lateral anterior orbitotomy aiming to free up the cyst as far as possible before approaching the probable rupture point at the dura attachment superiorly. The last surgical step was then performed through an anterior approach, the most vulnerable point for the rupture at the level of the

dura connection superiorly. At this level, the cyst was fully attached to the dura and had to be opened. Prior fine needle aspiration was not effective, as the needle has been blocked and a wider needle diameter caused a splitting of the cyst, spilling some content. Therefore, the cyst was finally emptied by suction and then fully removed (► **Fig. 1 b**).

The postoperative anthelmintic therapy (praziquantel 3000 mg every 24 hours for 1 week, albendazole 300 mg every 12 hours for > 10 months) initially caused severe vomiting and heart pain and had to be reduced until it was well tolerated. Amoxicillin and clavulanic acid were added due to a post-inflammatory reaction and fever.

Five months later, the swelling disappeared completely, the extraocular muscles retracted, and the levator muscle continuously improves its activity. Further reconstructive corrections such as ptosis surgery and repositioning of the globe may be discussed in a few months, when at least 1 year after surgery will have passed.

Discussion

In summary, determining the perfect time to perform surgery in a pregnant patient with orbital CE is demanding. Pre-/peri- and postsurgical side effects of surgery (including the high risk of cyst rupture) and anthelmintic medication might justify performing surgery and treatment after childbirth in case of an already lost eye in order to avoid risks for the mother and unborn child. In our patient, the spontaneous childbirth did not cause any rupture of the cyst.

Further case reports investigating this topic as well as the management of pregnant women with a good visual prognosis are needed.

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Conflict of Interest

The authors declare that they have no conflict of interest.

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References

- [1] Lentzsch AM, Göbel H, Heindl LM. Primary Orbital Hydatid Cyst. *Ophthalmology* 2016; 123: 1410. doi:10.1016/j.ophtha.2016.02.042
- [2] Thapa S, Ghosh A, Ghartimagar D et al. Hydatidosis of infratemporal fossa with proptosis – an unusual presentation: a case report and review of the literature. *J Med Case Rep* 2018; 12: 309. doi:10.1186/s13256-018-1812-y
- [3] Assimakopoulos SF, Marangos M. Orbital Hydatid Cyst. *N Engl J Med* 2020; 382: 1352. doi:10.1056/NEJMicm1911903

- [4] Chtira K, Benantar L, Aitlhaj H et al. The surgery of intra-orbital hydatid cyst: a case report and literature review. *Pan Afr Med J* 2019; 33: 167. doi:10.11604/pamj.2019.33.167.18277
- [5] Fasina O, Ogun OG. Hydatid cyst of the orbit in a young Nigerian female: a case report. *Ghana Med J* 2017; 51: 204–206
- [6] Oztekin PS, Yilmaz BK, Gokharman FD et al. Primary orbital hydatid cyst: computed tomography and magnetic resonance imaging findings. *Singapore Med J* 2014; 55: e184–e186. doi:10.11622/smedj.2014167
- [7] Dziri C, Haouet K, Fingerhut A et al. Management of cystic echinococcosis complications and dissemination: where is the evidence? *World J Surg* 2009; 33: 1266–1273. doi:10.1007/s00268-009-9982-9
- [8] Minciullo PL, Cascio A, David A et al. Anaphylaxis caused by helminths: review of the literature. *Eur Rev Med Pharmacol Sci* 2012; 16: 1513–1518

Bibliography

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