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Abstract

Case summary A 6-week-old entire female domestic shorthair cat was presented for evaluation of a soft bulge and a palpable skull defect on the forehead, present since adoption a few days earlier. The neurological examination revealed an absent menace response bilaterally and apparent blindness, localising the lesion to the occipital cortex. The main differential diagnoses were meningocele (MC) and meningoencephalocoele (MEC). Surgical repair was proposed once the cat reached adult size. Meanwhile, the cat developed seizures and was treated with anticonvulsant therapy. At 6 months of age, CT confirmed a frontoparietal MEC with associated porencephaly. Based on a three-dimensional printed skull mould, a polysulfone implant was created. The meninges were dissected from the skin, a durectomy was performed and samples of the protruding brain were obtained. Part of the cerebrospinal fluid was drained until the size of the protruding brain decreased enough to be included below the implant that was anchored on top of the skull with cerclages. Histopathology confirmed the diagnosis of MEC. Three years and 7 months later, the cat had partially recovered vision but continued to seize monthly despite antiepileptic drugs.

Relevance and novel information MC/MEC is a relatively uncommon disease reported in companion animals, and only four cases of surgical management have been described, and did not use a polysulfone tailor-made implant. In human medicine, surgical intervention is the treatment of choice. This case highlights a new implant option for surgical correction of MEC with good long-term result and no complications after 3 years and 7 months.

Keywords: Meningoencephalocoele; polysulfone tailor-made implant; 3D reconstruction; frontoparietal

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Introduction

Meningoencephalocoele (MEC) refers to a focal failure of the neuroectoderm and surface ectoderm to separate during embryonic development. This induces a protrusion of meninges and cerebral tissue through an opening in the calvaria (cranioschisis or cranium bifidum) that might be covered by skin.¹ Numerous factors have been suggested as potential causes for this malformation, including genetic defects, exposure to teratogenic agents during pregnancy and nutritional deficiencies.^{1–4}

Meningocele (MC)/MEC is a relatively uncommon disease reported in companion animals,^{1–12} and only four cases of surgical correction have been described.^{1,8,10,11} Although a multicentric retrospective study involving

22 dogs suggested that medical management could be an option for dogs with MC/MEC and mild neurological signs,² surgical intervention is considered the treatment of choice in human medicine.¹³

The objective of this clinical report is to describe the use of a tailor-made polysulfone implant for the surgical

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correction of an extensive frontoparietal MEC in an 8-month-old cat and the long-term results.

Case description

A 6-week-old entire female domestic shorthair cat was presented for evaluation of a soft bulge and a palpable skull defect on the forehead since adoption a few days earlier (Figure 1). The physical examination did not reveal any other abnormalities. The only neurological deficit was an absent menace response bilaterally, which was attributed to the young age of the cat and the apparent blindness (see the video in the supplementary material), localising the lesion to the occipital cortex. Differential diagnoses included congenital or traumatic MC or MEC. Echography through the cranial defect showed that the protruding tissue contained cerebral cortex and was associated with porencephaly. Therefore, a diagnosis of MEC and porencephaly was achieved. Surgical repair when the cat reached adult size was proposed.

At the age of 4 months, the cat developed generalised tonic-clonic seizures and was initially treated with levitiracetam (20 mg/kg q8h PO gradually increased to 40 mg/kg q8h PO owing to poor seizure control [Keppra oral solution 100 mg/ml; UCB Pharma SA]). Then, when the cat was 6 months old, phenobarbitone (2 mg/kg q12h PO gradually increased to 7.5 mg/kg q12h PO to reach an acceptable serum level and seizure control [Luminaletas 15 mg; Kern Pharma]) was added. At that stage, a CT study was performed under general anaesthesia with a 16-slice helical CT scanner (Brivo CT385; General Electric Healthcare). The CT images confirmed the presence of severe cranioschisis with complete absence of the frontal bone, including the frontal sinuses and part of the parietal bone. Both the meninges and cortical tissue protruded through the defect and porencephaly was also present. The caudal cranial fossa was subjectively small, and the cerebellum appeared flattened dorsoventrally (Figure 2). A three-dimensional printed reconstruction of the cat's skull helped to adapt a polysulfone tailor-made implant (Beta Implants; Pontevedra) for a perfect fit to cover the bone defect (Figure 3).

Surgical resolution was scheduled for 2 months later when the cat was 8 months old. Intramuscular pethidine (5 mg/kg IM [Dolantina 100 mg/2 ml; Kern Pharma]) and alfaxalone (0.5 mg/kg IM [Alfaxan 10 mg/ml; Dechra]) were used as premedication. Induction was carried out with alfaxalone (0.5 mg/kg IV) and midazolam (0.2 mg/kg IV [Midazolam 15 mg/3 ml; Normon]), and an alfaxalone continuous rate infusion (CRI) was used for anaesthetic maintenance at 10 mg/kg/h. During surgery, a fentanyl (Fentanest Kern Pharma) CRI was administered at 5 µg/kg/h and cefazolin (Cefazolin Normon) at 20 mg/kg every 1.5 h. No anaesthetic complications were noted during the surgery.



Figure 1 Preoperative image of the cat at presentation showing the large bulge in the forehead

For the surgical intervention, the cat was aseptically prepared. The dorsal frontal region of the head was shaved, from the dorsal limit of the nose to the second cervical vertebra, including the lateral canthus of both eyes and the ears. The area was disinfected with 1% povidone-iodine (Braunosan H Plus Braun). The cat was positioned in straight sternal recumbency, with the head placed on a sandbag and firmly fixed to the table. A longitudinal incision was performed along the malformation midline and extended 2 cm rostrally and caudally. The subcutaneous tissue was then carefully dissected to separate the skin from the abnormally thickened duramater, and the MEC was isolated. Intravenous mannitol (0.25 g/kg [Mannitol 20%; Braun]) and dexamethasone (0.05 mg/kg IV [Caliercortin 4 mg/ml Calier]) were administered as preventive treatment in case a subclinical increased intracranial pressure was present, before performing the durectomy to remove the abnormal meninges and expose the protruded cerebral cortex, whose macroscopic appearance was also abnormal. The resected meninges were sent for histopathological analysis. The exposed brain was hard and thickened, and no gyri were identifiable (Figure 4a). A 2 × 2 mm wedge biopsy of the protruding brain was also submitted for histopathological study. Part of the cerebrospinal fluid lodged in the porencephalic cavity was drained with a 1 ml syringe until the volume of the protruding brain decreased enough to be enclosed by the tailor-made implant. Four holes were drilled in the implant and another four at the edges of the cranial defect using a 1.5 mm drill bit so that they coincided

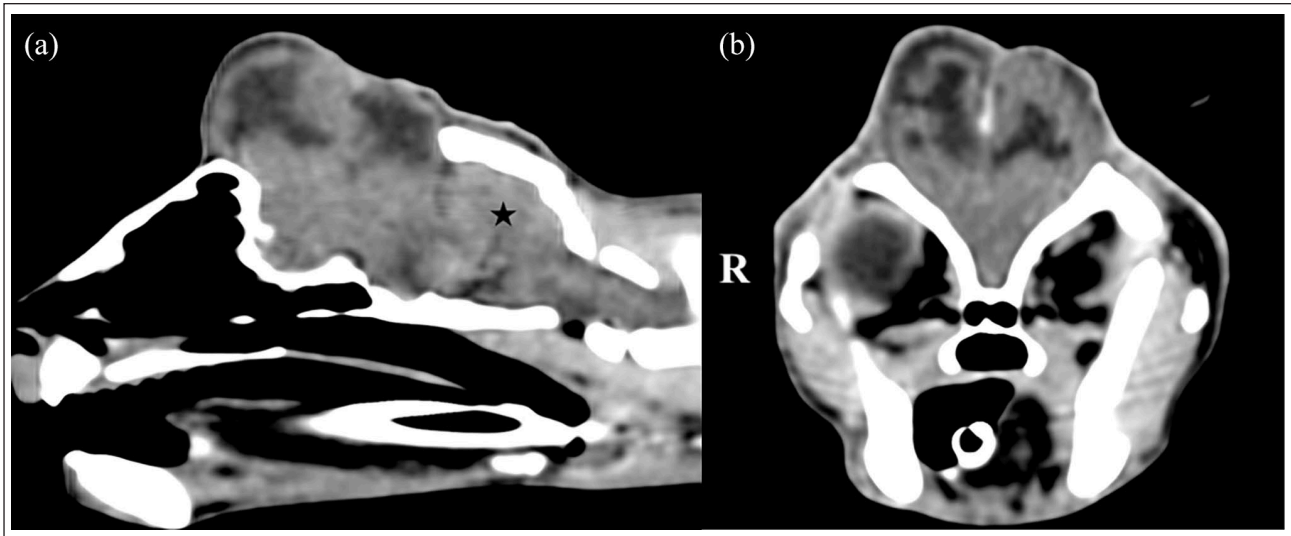


Figure 2 (a) CT mid-sagittal reconstruction soft tissue algorithm showing severe cranioschisis with complete absence of the frontal bone, including the frontal sinuses and both meninges and brain tissue protruding through the defect consistent with meningoencephalocele. Note a subjectively smaller-than-normal caudal cranial fossa and flattening of the cerebellum (black star); and (b) CT transverse plain of the same images at the level of the frontal lobe

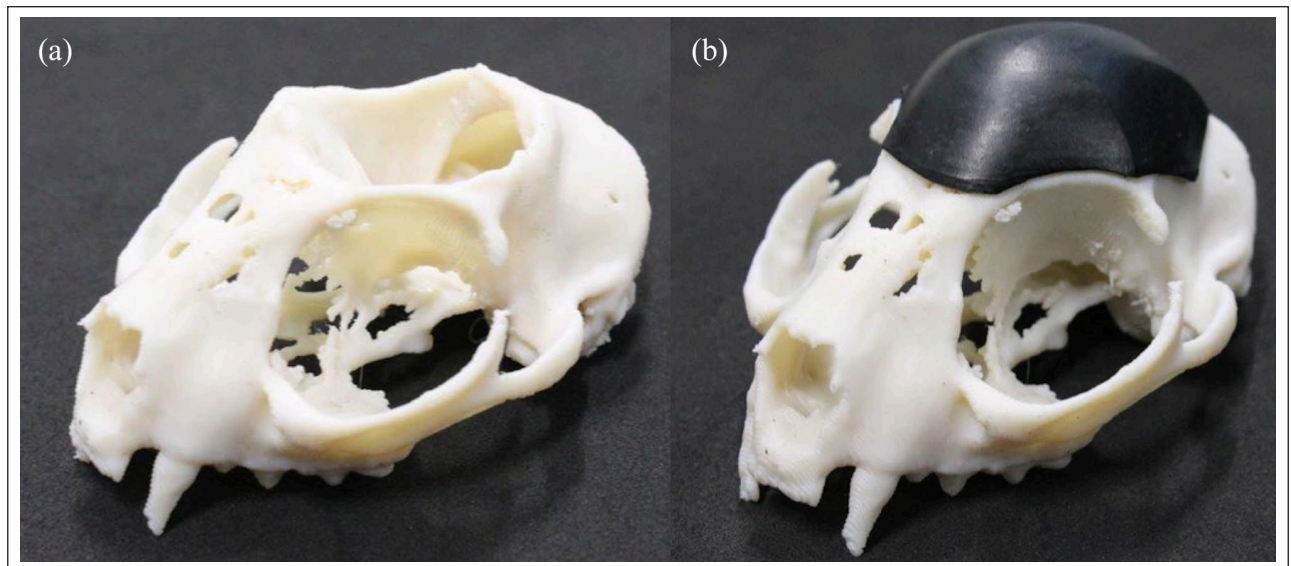


Figure 3 (a) Three-dimensional printed skull mould of the cranium of the cat at 6 months of age; and (b) the tailor-made polysulfone implant

once the implant had been placed in the desired position. A haemostatic collagen mesh (Lyostyp; Braun) was placed between the brain tissue and the implant to replace the duramater. The implant was anchored to the skull with four 0.6 mm cerclages in the predrilled holes (Figure 4b). The skin excess was removed using Metzembaum scissors to achieve a cosmetically adequate appearance (Figure 5). The skin closure was performed in layers.

A postoperative CT revealed an acceptable congruence between the implant and the skull with minimal pneumocephalus (Figure 6).

Postoperative treatment included cefazolin (20 mg/kg q8h IV), dexamethasone (0.05 mg/kg q24h IV), phenobarbitone (6.5 mg/kg q12h IV), levetiracetam (30 mg/kg q8h IV), maropitant (1 mg/kg q24h IV [Cerenia Zoetis]) and a fentanyl CRI at 3 µg/kg/h, which was progressively reduced over the first 24h and replaced by buprenorphine (10 µg/kg q8h IV [Buprecare Divasa-Farmavic SA DFV Group]). The cat was conscious and ambulatory within 12h of surgery but showed ataxia and mild obtundation, which gradually improved during hospitalisation. The cat was discharged 3 days later with slight obtundation, mild ataxia and bilateral blindness.

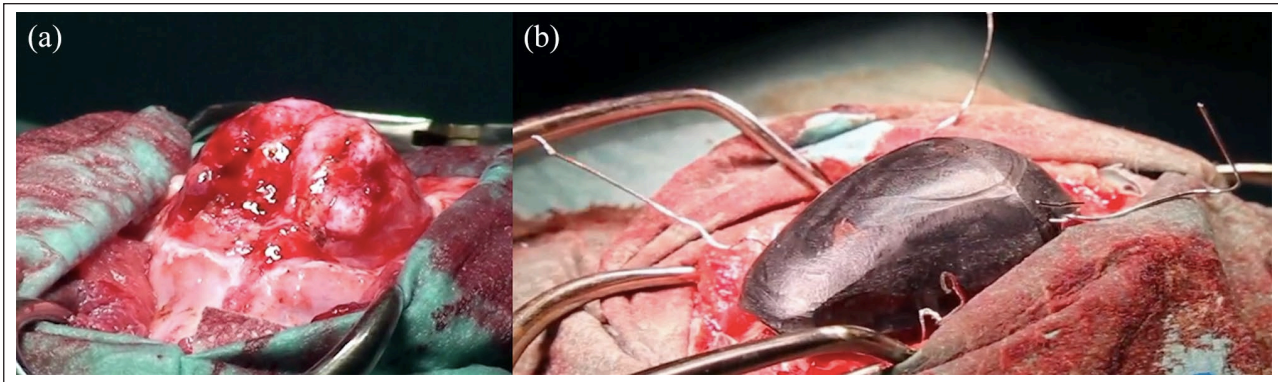


Figure 4 (a) Intraoperative image showing the exposed protruding brain tissue after craniotomy; and (b) intraoperative image showing the implant placement and fixation with cerclages within predrilled holes



Figure 5 Postoperative image of the cat a few months after surgery, showing acceptable cosmetic results

The histopathological and immunohistochemical results showed disorganised nervous tissue and leptomeningeal thickness. The cerebral cortex was deprived of gyri and showed abnormal neural architecture with loss of columnar and laminar organisation, a marked decrease in the number of neuronal cell bodies and microspangiosis of the neuropil. Areas of neuronal pyknosis were observed along with reactive gliosis, astrogliosis, a large number of rod cells and activated macrophages containing cytoplasmic dark material. The subcortical white matter showed loss of eosinophilia and excessive nerve fibre separation. The leptomeninges were accompanied by vascular proliferation and an increase in the fibrous collagen-like component. The duramater was disorganised, with abundant dense collagen bundles and calcification. Inflammation was not a feature (Figure 7). These findings were all consistent with an MEC.

Check-ups were carried out 1 week after surgery, 3 weeks later, then 2 months and 6 months after the intervention. At that time, the cat had partially recovered vision despite an absent menace response bilaterally. Otherwise, the neurological examination was normal, and the cat had no seizures since the surgery 6 months earlier. However, the cat started seizing again when a gradual decrease in levetiracetam was initiated and the treatment was reinstated.

One year after surgery, the cat was admitted for a conventional ovariohysterectomy that proceeded without complications. At that point, a recheck CT was performed observing that the implant was still in the same position and the pneumocephalus, present in the CT immediately postsurgery, had resolved (Figure 8).

Three years and 7 months after surgery, the cat remained stable with a good quality of life and partial visual capacity, despite bilaterally absent menace response. At the time of final follow-up, 43 months postoperatively, the cat was seizing monthly despite receiving phenobarbitone (6.5 mg/kg q12h) and levetiracetam (30.5 mg/kg q8h).

Discussion

In human medicine, surgical intervention is considered the treatment of choice for MEC.¹³ This is based on the potential for recurrent meningitis, brain damage from herniation and refractory seizures, as MECs are thought to act as seizure foci.² Goals include removing the dural sac, preserving functional brain tissue and obtaining adequate wound closure.¹

Although MEC/MCs have been described in dogs and cats,^{1–12} and also occasionally reported in other species, including calves,^{14–18} pigs,¹⁹ a rabbit²⁰ and a raccoon,²¹ most of those animals were euthanased or treated conservatively. Medical management has been suggested for 22 dogs with MC/MEC and mild neurological signs.² However, 73% of those dogs suffered an

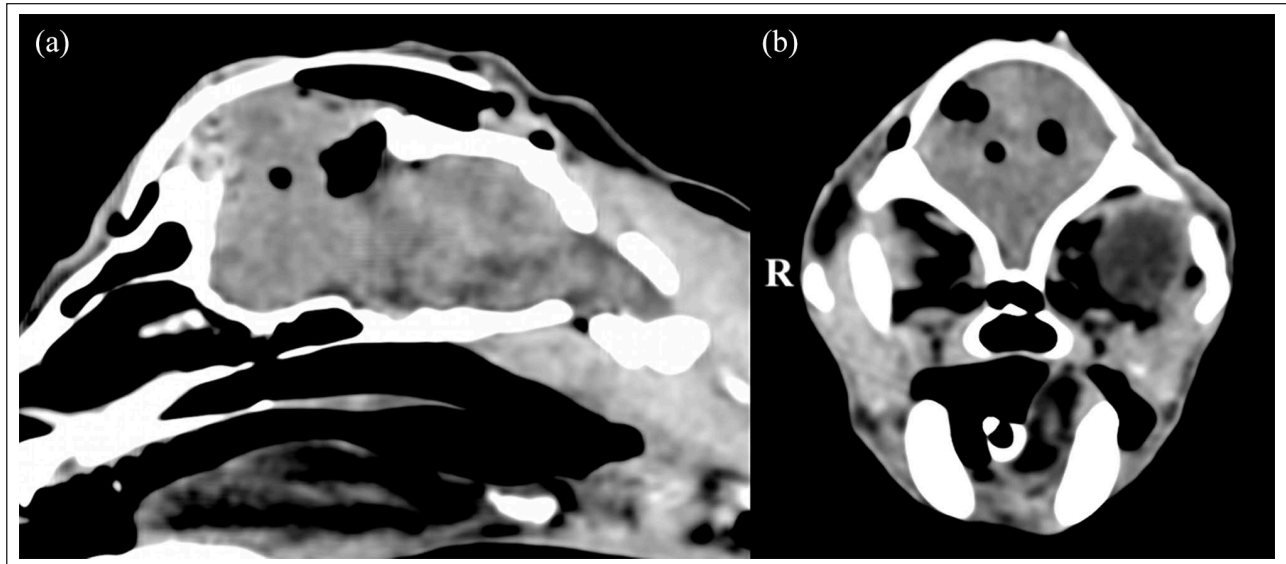


Figure 6 (a) Immediate postoperative CT mid-sagittal reconstruction soft tissue algorithm showing the tailor-made implant in place. Note the presence of postoperative pneumocephalus. (b) CT transverse plain of the same images at the level of the frontal lobe

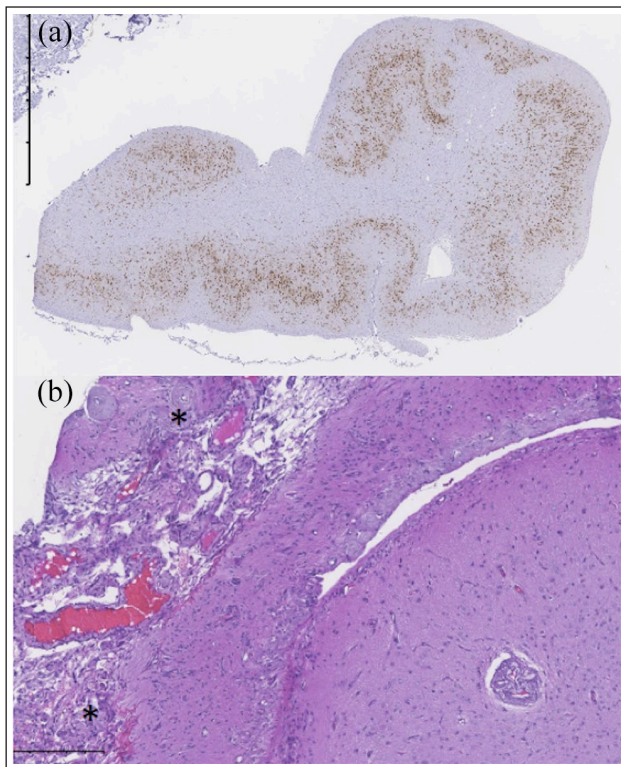


Figure 7 (a) Microscopic transverse section of the abnormal protruding cerebral cortex (NeuN immunohistochemistry counterstained with haematoxylin). Note the absence of sulci and abnormal organisation of neuronal laminae with irregular density of neuronal cell bodies. (b) Microscopic transverse section of the abnormal protruding tissues (haematoxylin and eosin staining). Note leptomeningeal thickening (*) and cell proliferation forming fibrous solid or concentric structures. The adjacent cortical cerebral tissue shows abnormal distribution of neuronal cell bodies

intranasal MEC and only 27% had the parietal bone affected; all of them with only meninges protruding through the parietal bone defect.² On the other hand, surgical correction has only been described in two dogs,^{8,10} two cats^{1,11} and two calves.^{15,17} The scarce number of MEC cases described in the veterinary literature precludes any conclusion regarding the best treatment option for this pathology, and needs to be evaluated on a case-by-case basis. Given the size of the bone defect and the large amount of protruding neural tissue in the present case, surgical repair was considered the best treatment for this cat.

Both surgically treated dogs described previously,^{8,10} and one of the cats,¹¹ suffered from an ethmoidal MEC and seizures, which were repaired after resection of the herniated tissue. In one of these dogs⁸ and the cat,¹¹ seizures resolved after surgical resection; however, seizure frequency did not change after surgery in the other dog,¹⁰ which was attributed to the development of additional seizure foci as the dog had been seizing 36 months prior to the surgery. Finally, the sole cat reported with a parietal MEC had only a small amount of protruded brain tissue, and an abrupt resolution of aggressiveness was also observed after surgical resection of the herniated tissue.¹ The present case represents the largest surgically treated MEC; a complete resection was not considered as it might have affected the cat's quality of life. This could have contributed to the fact that the cat continued seizing after surgery; however, both concurrent malformations (porencephaly and an abnormally small cranial caudal fossa present in our case) and a large volume of herniated neural tissue have also been described as negative prognostic factors in human medicine.¹³

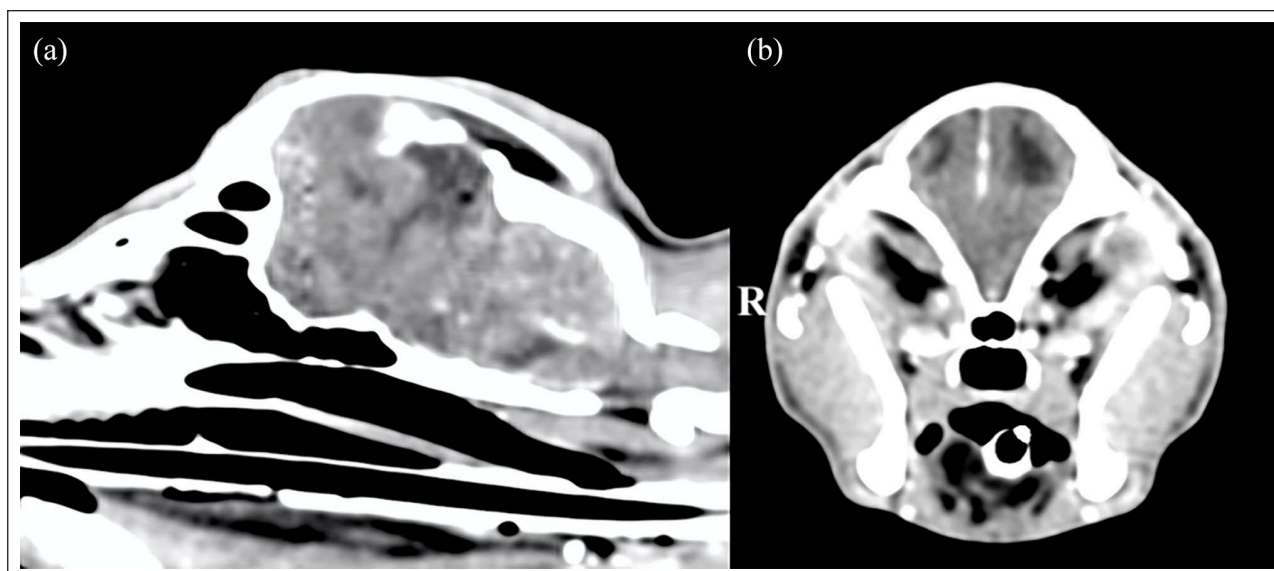


Figure 8 (a) Twelve-month postoperative CT mid-sagittal reconstruction soft tissue algorithm showing complete resolution of the postoperative pneumocephalus and correct implant positioning. (b) CT transverse plain of the same images at the level of the frontal lobe

Regarding the different implant options available to close the MEC bone defect, described techniques in companion animals include dural substitutes and/or autologous fascia,^{8,10,11} and a titanium mesh covered by polymethylmethacrylate on both sides.¹ Dural substitutes and/or autologous fascia have only been used to repair ethmoidal MEC;^{8,10,11} they were not considered in the present case owing to the location and extension of the cranial defect. A hard implant was required to protect the brain below. Comparing the implant used in our case with the titanium mesh covered by polymethylmethacrylate,¹ the latter is more voluminous as it requires a greater thickness to achieve the same resistance and rigidity; it is heavier (4.42 g/cm³ vs 1.31 g/cm³); more difficult to adapt intraoperatively; and it is radiopaque, so it could interfere in future advanced imaging results. The tailor-made polysulfone implant was chosen for several reasons. First, given the extension of the defect, we needed a more dome-shaped implant than commonly used, which would have been more difficult to achieve with the titanium mesh and polymethylmethacrylate. Secondly, being able to tailor it before the surgery allowed the implant to perfectly fit in the defect. Finally, having the implant ready at the time of surgery allowed us to save surgical time that we would have spent adjusting the titanium mesh to the defect and covering it with polymethylmethacrylate on both sides.

For the tailor-made implant design, a specific software for industrial three-dimensional design was used. First, a virtual three-dimensional model to simulate the surgery was generated from the CT images and the surgeon's indications. The implant was then created to fulfil adequate adaptation to the skull anatomy, physical

and mechanical properties, volume to hold the tissue and good aesthetic results. Recently, three-dimensional printing has become increasingly popular in the veterinary community as models for teaching purposes,^{22–24} and also in clinical neurosurgery as three-dimensionally printed titanium plates for cranioplasty in canine skull tumours;²⁵ three-dimensionally printed patient-specific drill guides for spinal deformities,²⁶ vertebral fractures²⁷ and atlanto-axial ventral stabilisation;²⁸ and imaging-based three-dimensionally printed stereotactic brain biopsy devices in dogs.²⁹

Conclusions

To our knowledge, the case described here is the first to use a tailor-made polysulfone implant based on a three-dimensionally printed mould to close an MEC defect, and provided excellent cosmetic results. This case highlights a new implant option for surgical correction of MEC with good long-term results and no complications to date.

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Supplementary material The following file is available online:

Video 1: Neurological examination of the kitten at 6 weeks.

Conflict of interest The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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Ethical approval The work described in this manuscript involved the use of non-experimental (owned or unowned) animals. Established internationally recognised high standards ('best practice') of veterinary clinical care for the individual patient were always followed and/or this work involved the use of cadavers. Ethical approval from a committee was therefore not specifically required for publication in *JFMS Open Reports*. Although not required, where ethical approval was still obtained, it is stated in the manuscript.

Informed consent Informed consent (verbal or written) was obtained from the owner or legal custodian of all animal(s) described in this work (experimental or non-experimental animals, including cadavers) for all procedure(s) undertaken (prospective or retrospective studies). For any animals or people individually identifiable within this publication, informed consent (verbal or written) for their use in the publication was obtained from the people involved.

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