

Balloon dilation and transient stenting of unilateral membranous choanal atresia in a British Shorthair cat with chronic purulent rhinitis and ascending meningoencephalitis

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Ballondilatation und vorübergehende Stentimplantation bei einer einseitigen membranösen Choanalatresie bei einer Britisch Kurzhaarkatze mit chronischer eitriger Rhinitis und aufsteigender Meningoenzephalitis

Choanalatresie ist eine seltene angeborene Anomalie bei Menschen und Tieren, die durch die fehlende Verbindung einer oder beider Nasenhöhlen mit dem Nasopharynx gekennzeichnet ist. Die Schwere der klinischen Symptome hängt vom Vorliegen einer einseitigen oder beidseitigen Stenose sowie von Komorbiditäten ab. Bei einer bilateralen Atresie kann die Atmung insbesondere im Schlaf stark beeinträchtigt sein, da der Luftstrom nur bei der Atmung durch den offenen Mund erfolgen kann. Verschiedene Therapiemodalitäten wurden beim Menschen beschrieben und für Tiere übernommen. Bei allen Behandlungen können Komplikationen auftreten, am wichtigsten ist die posttherapeutische Narbenbildung mit erneuter Stenose. Dieser Bericht beschreibt eine 10 Monate alte Britisch Kurzhaarkatze mit chronischem einseitigem serösem Nasenausfluss, der sich schliesslich zu einem mukopurulenten Ausfluss entwickelte. Als akute neurologische Symptome auftraten, wurde die Katze in der Klinik vorgestellt. Die Diagnose einer primären, membranösen rechtsseitigen Choanalatresie wurde mittels Computertomographie und nasopharyngealer Rhinoskopie (von hinten) gestellt. Zu den sekundären Veränderungen gehörten eine destruktive Rhinitis mit Progression in das ZNS mit einem subduralen Empyem und einer Meningoenzephalitis. Netzhautveränderungen und eine Aspirationspneumonie wurden ebenfalls als Komplikationen beurteilt. Nach der klinischen Abheilung der Sekundärinfektionen wurde die membranöse Obstruktion perforiert und mittels eines Ballonkatheters aus der Kardiologie durch einen orthograden transnasalen Zugang unter endoskopischer Führung aus retroflexierter nasopharyngealer Sicht erweitert. Zur Ver-

Summary

Choanal atresia is a rare congenital anomaly in humans and animals, characterized by the absence of communication of one or both nasal cavities with the nasopharynx. The severity of clinical signs depends on the presence of unilateral versus bilateral stenosis as well as comorbidities. With bilateral atresia, respiration may be severely compromised particularly during sleep, as airflow can only occur when breathing through the open mouth. Various therapeutic modalities have been described in people and adopted for animals. All treatments may be associated with complications, the most important being post-therapeutic scar formation with re-stenosis. This report describes a 10-month-old British Shorthair cat with chronic unilateral serosal nasal discharge that changed to mucopurulent discharge. When acute neurological signs developed, the cat was presented to the veterinary hospital. A diagnosis of primary, membranous right sided choanal atresia was achieved via computed tomography (CT) and nasopharyngeal (posterior) rhinoscopy. Secondary changes included destructive rhinitis with progression to the CNS with a subdural empyema and meningoencephalitis. Retinal changes and aspiration bronchopneumonia were suspected additional complications. After recovery from the secondary infections, the membranous obstruction was perforated and dilated using a valvuloplasty balloon by an orthograde transnasal approach under endoscopic guidance from a retroflexed nasopharyngeal view. To prevent re-stenosis, a foley catheter was placed as a transient stent for 6 days. The cat recovered uneventfully and was asymptomatic after the stent removal. Endoscopic re-examination after 5 months confirmed a persistent opening and patency of the generated right choanal passage. The cat remains asymptomatic 10 months after the procedure. Transnasal endoscopic balloon dilation and transient stenting of choanal atresia is a minimally invasive and relatively simple procedure with potentially sustained success.

Keywords: cat, subdural empyema, valvuloplasty, nasopharyngeal stenosis, foley catheter

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hinderung einer Restenose wurde für 6 Tage ein Foley-Katheter als vorübergehender Stent platziert. Die Katze erholte sich ereignislos und war nach Entfernung des Stents asymptatisch. Eine erneute endoskopische Untersuchung nach 5 Monaten bestätigte eine anhaltende Öffnung und Durchgängigkeit des erzeugten rechten Choanalgangs. Die Katze bleibt auch 10 Monate nach dem Eingriff asymptatisch. Die transnasale endoskopische Ballondilatation und die vorübergehende Stentimplantation einer Choanalatresie ist ein minimalinvasiver und relativ einfacher Eingriff mit potenziell nachhaltigem Erfolg.

Schlüsselwörter: Katze, Subdural Empyem, Ballondilatation, Nasopharynxstenose, Foley-Katheter

Case report

A 10-month-old, male castrated British Shorthair cat (body weight 4kg) was presented for further investigation of chronic unilateral serosal nasal discharge with acute deterioration into mucopurulent discharge and acute onset of neurological signs. The owner reported serosal unilateral

nasal discharge since the cat was acquired at the age of 3 months. When quantity and quality of the discharge progressively worsened, the cat had been presented to a referring veterinarian at the age of 5 months. Rhinoscopic examination revealed a «friable membrane» in the right nasal cavity, which reportedly was perforated with the endoscope, and the cat subsequently was treated with antibiotics. No improvement was noted. At the age of 10 months, the cat was referred due to severe unilateral mucopurulent nasal discharge, severe lethargy, anisocoria and blindness.

At presentation, the cat was stuporous, mildly dehydrated, tachypneic (respiratory rate 60/min) and subfebrile (rectal temperature 39,1°C). There was severe mucopurulent right-sided nasal discharge and the mandibular lymph nodes were mildly enlarged bilaterally. The neurological examination showed decreased proprioception in both forelimbs and an absent pupillary light reflex and menace response on the right side. Ophthalmological changes consisted of right-sided blepharospasm with mild anisocoria, prolapse of the 3rd eyelid and bilateral retinal oedema. Blood pressure measurement (Suntech, cuff 2, left forearm) revealed mild hypertension (166/124/134 mmHg). Thoracic radiographs showed several areas of increased radiodensity confluent to zones with an alveolar lung pattern. Laboratory abnormalities included lymphocytosis ($7,7 \times 10^3/\mu\text{L}$, reference 1,0– $6,0 \times 10^3/\mu\text{L}$), mildly increased ASAT (125 U/L, reference 19–44 U/L), moderately increased CK (5007 U/L, reference <355 U/L), and moderately increased serum amyloid A (163,4 mg/dL, reference <3,9 mg/dL). Fasted bile acids were normal (2,3 μmol/L, reference <6,5 μmol/L). A PCR for feline Herpes- and Calicivirus, collected by an oropharyngeal swab, was negative. Initial treatment consisted of intravenous fluid (plasmalyte 3 ml/kg/h), opioid (butorphanol 0,2 mg/kg i.v.) and antibiotic therapy (amoxicillin-clavulanic acid 20 mg/kg i.v. q8h) as well as eye ointment into the right eye (oxytetracycline q8h).

To further examine nose and brain, a computed tomography (CT) of the head was performed under general anaesthesia.

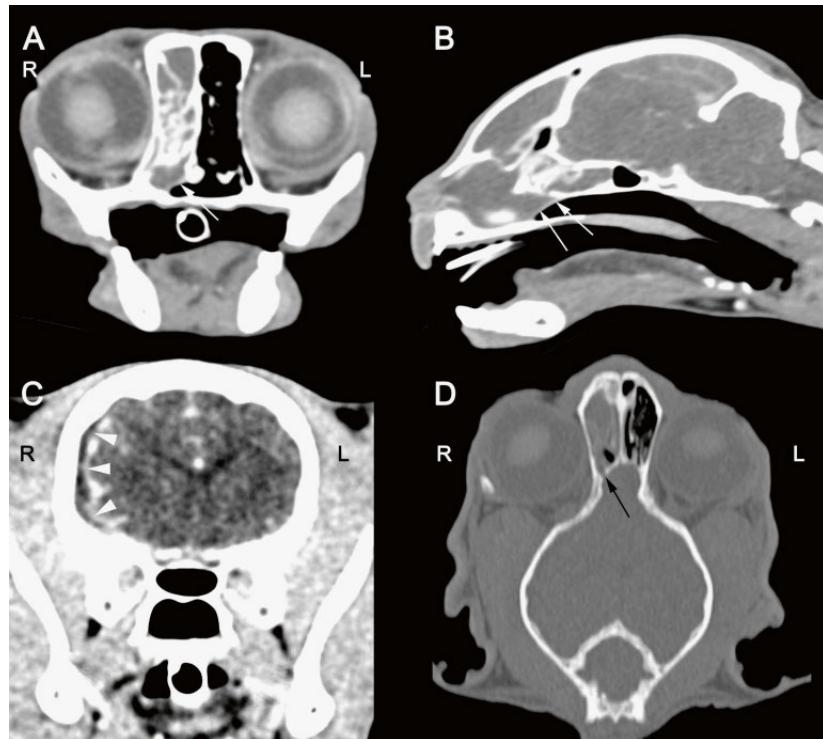


Figure 1: Reconstructions of the computed tomography (CT) examination of the head of a 10-month-old British Shorthair kitten with nasal discharge and central nervous system deficits. A) Transverse and B) sagittal soft tissue reconstruction in a soft tissue window, C) transverse soft tissue reconstruction, windowing optimized for the visualization of the meningeal splitting, D) dorsal reconstruction in a bone reconstruction and bone windowing. A soft tissue band (white arrows) divides the right nasal cavity in two compartments with accumulation of mucus dorsally and a narrowed passage ventrally (figure 1A & 1B). Severe meningeal splitting with subdural accumulation of fluid (white arrow heads) and thickening of the enhancing meninges (figure 1C). Small discontinuity in the cribriform plate (black arrow) (figure 1D).

On the CT images the right nasal cavity was completely filled with non-enhancing fluid attenuating material. This material extended to and filled the right frontal sinus and the right sphenoidal sinus. The lesion was associated with osteolysis of the cribriform plate at its right dorsolateral aspect and extended intracranially (extra-axially) widening the subdural space and displacing the right frontal cortex, causing a mild leftward midline shift. The adjacent menin-

ges, suspected to be the pachymeninges, showed a markedly increased contrast uptake (figure 1A). The mandibular and medial retropharyngeal lymph nodes were moderately enlarged. Finally, there was a thin and well-defined, soft tissue attenuating band between the right ventral conchal meatus and the choana, impairing the communication with the nasopharynx (figure 1B & C).

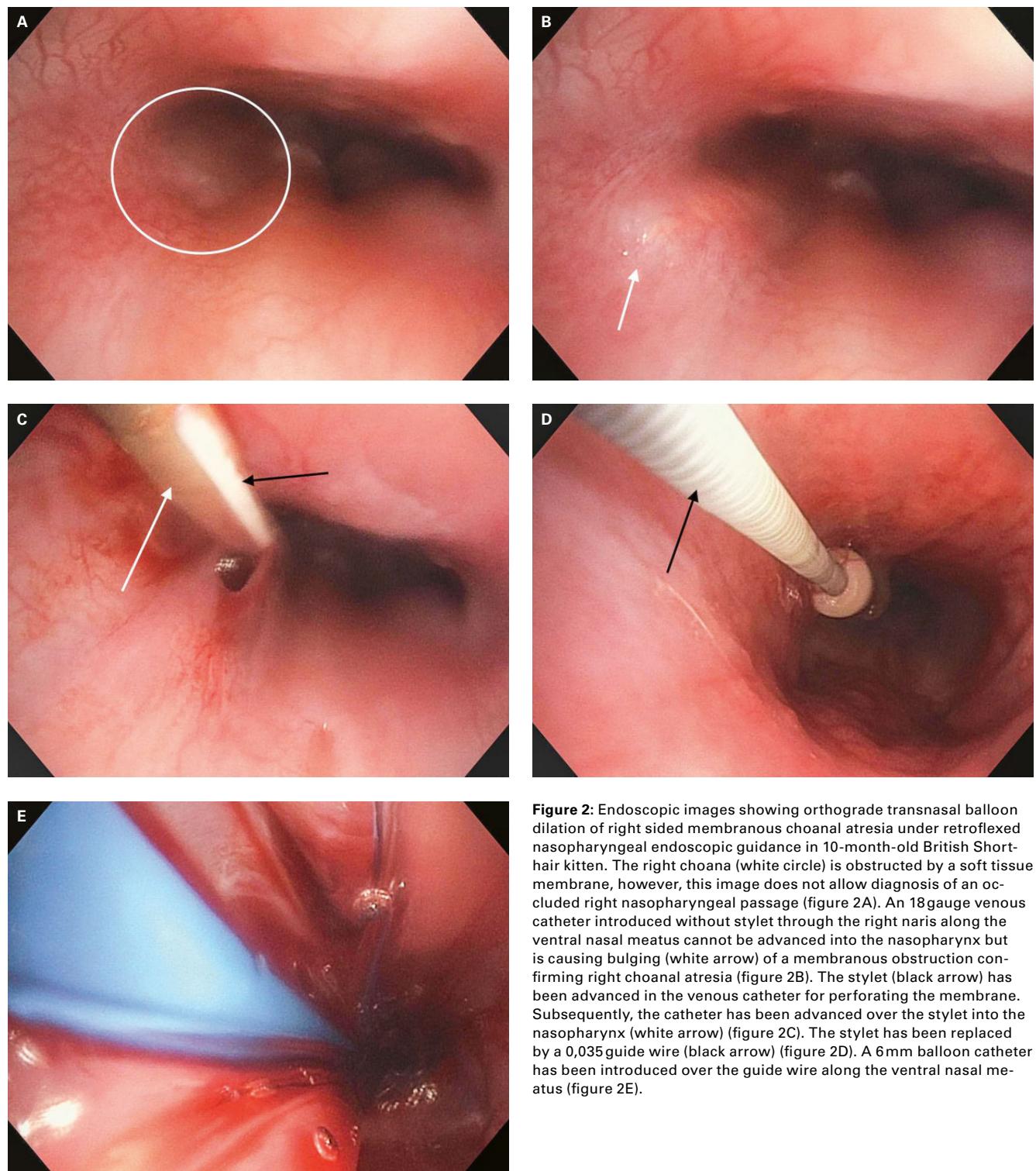


Figure 2: Endoscopic images showing orthograde transnasal balloon dilation of right sided membranous choanal atresia under retroflexed nasopharyngeal endoscopic guidance in 10-month-old British Short-hair kitten. The right choana (white circle) is obstructed by a soft tissue membrane, however, this image does not allow diagnosis of an occluded right nasopharyngeal passage (figure 2A). An 18gauge venous catheter introduced without stylet through the right naris along the ventral nasal meatus cannot be advanced into the nasopharynx but is causing bulging (white arrow) of a membranous obstruction confirming right choanal atresia (figure 2B). The stylet (black arrow) has been advanced in the venous catheter for perforating the membrane. Subsequently, the catheter has been advanced over the stylet into the nasopharynx (white arrow) (figure 2C). The stylet has been replaced by a 0,035 guide wire (black arrow) (figure 2D). A 6mm balloon catheter has been introduced over the guide wire along the ventral nasal meatus (figure 2E).

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Anterior and posterior rhinoscopy showed severe accumulation of mucopurulent discharge. After collecting a sample for bacteriological examination from the nasal cavity, which revealed a mild growth of *Streptococcus pneumoniae*, the nasal cavity was flushed with saline solutions for better visualization of the choanal region. Right-sided, membranous choanal atresia (CA) was confirmed (figure 2A & B). Analysis of a CSF tap showed a low-grade mixed-cell pleocytosis with predominance of lymphocytes, but no infectious agents.

Based on these results, the diagnoses were 1) congenital membranous right sided CA, 2) secondary purulent rhinitis and 3) suspected aspiration pneumonia, 4) chorioretinitis, and 5) subdural empyema with bacterial meningoencephalitis.

After uneventful recovery from anaesthesia and before planning an intervention to correct the CA, suspected to be at the origin of all problems, the cat received antibiotic treatment (amoxicillin-clavulanic acid 20 mg/kg i.v. q 8h), intravenous fluids (plasmalyte 3 ml/kg/h i.v.), opioids (buprenorphine 15 mcg/kg i.v. q8h), and a NSAID (meloxicam 0,05 mg/kg p.o.).

The cat recovered quickly from the meningoencephalitis, chorioretinitis and pneumonia and was discharged after 3 days of hospitalization in good general condition; mild serosal to mucopurulent nasal discharge persisted. Oral antibiotic treatment was continued (amoxicillin-clavulanic acid 20 mg/kg p.o. q12h) until recheck.



Figure 3: Situation after transnasal balloon dilation of right sided membranous choanal atresia in 10-month-old British Shorthair kitten. After rupturing the obstructing membrane by balloon dilation there is a large opening (white arrow heads) without visible bleeding; the wire is still in place (figure 3A). Position of a 12 F Foley catheter placed for transiently stenting the created opening in order to avoid re-stenosis by scar tissue (figure 3B). External stent situation after recovery from anaesthesia (figure 3C).

The cat was re-evaluated after 8 weeks for treatment of the CA. At this time the only clinical abnormality was mild unilateral serosal nasal discharge. Under general anaesthesia, an over the needle catheter (18-gauge venous catheter) was introduced into the right naris and advanced along the ventral nasal meatus without the stylet to prevent damage to the nasal mucosa. Under endoscopic guidance from the nasopharyngeal retroflexed view, the catheter tip reached and tented the abnormal membrane (figure 2B). The stylet was now introduced and advanced to perforate the membrane, followed by advancement of the catheter across the membrane (figure 2C). The stylet was removed and a 0.035 inch guidewire introduced through the catheter into the nasopharynx (figure 2D). The catheter was then removed and a 6 Fr balloon catheter (Navigator II, Angiomed, PTA) was inserted (figure 2E), positioned and inflated several times for approximately 10 seconds each, which resulted in a large opening with only minimal bleeding (figure 3A). Finally, to avoid restenosis of the newly created opening by scar tissue, a 12 Fr Foley catheter was inserted along the right ventral nasal meatus across the opening, acting as a transient stent (figure 3B) and sutured via Chinese finger-trap to the nostril (figure 3C). The cat recovered uneventfully from anaesthesia and tolerated the stent very well. The cat remained hospitalized during the planned 7-day stenting phase. Amoxicillin-clavulanic acid was continued for 3 days and prednisolone was administered (1 mg/kg q24h for 3 days, then 0.5 mg/kg q24h). On day 6 after the procedure, the cat itself removed the stent.

A repeated rhinoscopic examination from the retroflexed nasopharyngeal view revealed no evidence of bleeding, inflammation or re-stenosis. The cat was discharged and prednisolone (0.5 mg/kg p.o. q24h) continued for 5 days.

Five months after the procedure, the cat was presented for a recheck. The owner reported a clinically normal cat without recurrence of any clinical signs and the cat had gained 500 g of bodyweight. By retroflexed nasopharyngoscopy, the size and patency of the generated right choana was re-evaluated. The size of the created opening was not visibly reduced since the intervention and patency was verified by inserting a feeding tube through the right ventral nasal meatus (figure 4). The cat received a single injection of dexamethasone (0.2 mg/kg i.v.) to prevent swelling secondary to mucosal irritation by the manipulations and was discharged the same day.

At a final follow-up by phone, 10 months after the catheter intervention and transient stenting, the cat continued to be clinically asymptomatic.

Discussion

Choanal atresia is a congenital craniofacial anomaly which prevents the communication of one or both nasal cavities with the nasopharynx, resulting in restricted or absent passage of airflow. In human medicine, this rare anomaly is more often unilateral than bilateral, and it has most commonly a mixed (71 %) or pure bony (29 %) rather than pure membranous structure.^{5,14} If the condition is bilateral, it can cause life-threatening dyspnea in newborns which may require emergency treatment in the neonate. Unilaterally affected patients are typically presented later, sometimes only as adults, with signs of unilateral obstruction and persistent nasal discharge.^{14,16,17,22} In people, CA can be isolated, but more commonly is associated with other major congenital defects, such as coloboma, heart abnormalities, growth or mental retardation, genitourinary anomalies, ear abnormalities, cranial nerve dysfunction, tracheo-oesophageal fistula and cleft lip/palate.^{12,16,22} In veterinary medicine, CA seems to be even less recognized than in people and reports in dogs and cats are scarce.^{2,7,13,15,23,24} One reason for the low recognition may be that severely symptomatic cases with life threatening dyspnea right after birth may never be presented to a veterinarian or be euthanized early in life. In cats, CA has only been described in 3 case reports, including 2 cats with unilateral, membranous and 1 cat with bilateral, bony atresia.^{2,15,24} A concurrent anomaly, hydrocephalus, was described in one cat.²

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Figure 4: Repeated nasopharyngeal endoscopy 5 months after transnasal rupture of right sided membranous choanal atresia. The created opening (white arrow heads) appears similarly open to the situation right after balloon dilation. Black arrow depicts tip of venous catheter introduced along ventral nasal meatus.

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Here we report an additional cat with unilateral, membranous CA, without any detectable concurrent congenital anomalies. However, a special complication in our cat was severe purulent rhinitis with ascending meningoencephalitis and choriorretinitis. The diagnosis of CA was obtained by CT of the head and subsequent posterior rhinoscopy.

A CT (or MRI) examination may not be necessary in every case, but may facilitate planning of subsequent treatment. Likewise, in the cat reported by Khoo et al. (2007) the diagnosis was made based on the findings upon nasopharyngoscopy alone.¹⁵ However, in human medicine a CT scan is highly recommended in order to confirm the diagnosis, plan the surgery, assess the nature and extent of the anomaly and not to miss concurrent congenital defects.¹⁹

Different surgical options to treat CA have been described in human medicine, including transnasal, transantral, transpalatal and transseptal techniques with different combinations of dilators, balloon dilation, microdebridement and drilling of the arrectic bony plates. The choice is based on the extend of the defect, concurrent anomalies and surgeon preferences.^{9,14,20,25}

In people, restenosis is the most common complication of repair and occurs in up to half of the cases. Therefore, multiple interventions are usually necessary.^{1,9} Use of postoperative stenting with silicone tubes has been recommended to avoid restenosis, and prognostic factors for success include a birth weight heavier than 2,3 kg, a duration of stenting longer than 12 weeks, and a stent size larger than 3,5 mm.^{10,14} However, several reports imply that outcome was not affected whether stents were used or not.^{1,9,19,25} Furthermore, using drugs to suppress scar formation like mitomycin were not statistically associated with the surgical result.¹⁹ In regards to clinical presentation, treatment options and outcome, congenital membranous CA is similar to acquired nasopharyngeal stenosis (NPS) and it has been suggested that some CA may actually rather be NPS.⁴ For the more common NPS, surgical as well as various techniques of balloon dilation with or without stenting and postoperative use of antibiotics and glucocorticoids have been described in cats, and mild to complete restenosis by scar tissue has been similarly reported irrespective of technique.^{3,4,11,18,21} In cases of severe re-stenosis, permanent stents may be placed, however, complications are common.⁶

In comparison, in the few reported cases of CA in small animal medicine, surgical techniques as well as balloon dilation have been used. In two dogs, rhinotomy was performed to surgically resect the stenotic abnormality.^{7,23} Both dogs developed restenosis by scar tissue, even though in one dog, in addition to surgery, a permanent stent had been placed. Further treatment in the first dog was a palliative tracheostomy⁷ and in the second dog, an additional stent was placed²³. In one cat with bilateral stenosis, surgical cor-

rection was performed first, and when restenosis occurred, ballooning was used followed by a stent.²⁴ In another two cats, ballooning of the stenosis with a foley catheter followed by stent placement for 7 and 15 days, respectively, was performed.^{2,15} Restenosis was not described in these latter two cases. In our case we used the same technique as we had described for balloon dilation of NPS¹¹, but we implanted a transient stent, similar to Khoo et al. (2007).¹⁵ At the moment of manuscript preparation, our cat shows no clinical signs of restenosis.

Indeed, as opposed to the reports in human medicine, transient stenting seems to be more promising for avoiding restenosis in cats⁸, and a transient stent avoids the long-term complications of a permanent stent.^{6,21} Nevertheless, while in the report by DeLorenzi et al. (2015)⁸, the outcome was described as most successful when using a stent after balloon dilation, this technique was not successful in the hands of Berent (2016).⁴ Therefore, it seems too early to draw any final conclusions, if stenting will markedly improve long-term outcome, and if the duration of transient stenting will affect the results.

In summary, this report provides further evidence that balloon dilatation of a congenital membranous CA followed by transient, short term stenting can lead to long-term success in cats.

Dilatation par ballonnet et pose d'une endoprothèse transitoire pour une atrésie choanale membranaire unilatérale chez un chat British Shorthair atteint d'une rhinite purulente chronique et d'une méningo-encéphalite ascendante

L'atrésie des choanes est une anomalie congénitale rare chez l'homme et l'animal, caractérisée par l'absence de communication d'une ou des deux cavités nasales avec le nasopharynx. La gravité des signes cliniques dépend de la présence d'une sténose unilatérale ou bilatérale, ainsi que des comorbidités. En cas d'atrézie bilatérale, la respiration peut être gravement compromise, en particulier pendant le sommeil, car l'air ne peut circuler que par la bouche ouverte. Diverses modalités thérapeutiques ont été décrites chez l'homme et adaptées pour les animaux. Tous les traitements peuvent être associés à des complications, la plus importante étant la formation de cicatrices post-thérapeutiques avec resténose. Ce rapport décrit un chat British Shorthair de 10 mois présentant un écoulement nasal séreux unilatéral chronique qui s'est finalement transformé en un écoulement muco-purulent. Lorsque des signes neurologiques aigus sont apparus, le chat a été présenté à l'hôpital vétérinaire. La tomodensitométrie (CT) et la rhinoscopie nasopharyngée (postérieure) ont permis de diagnostiquer une atrésie choanale primaire membranuse du côté droit. Les altéritiins secondaires comprenaient une rhinite destructrice avec une progression vers le SNC avec empyème sous-dural et méningo-encéphalite. Des altérations de la rétine et une bronchopneumonie par aspiration étaient des complications supplémentaires présumentes. Après guérison des infections secondaires, l'obstruction membranuse a été perforée et dilatée à l'aide d'un ballonnet de valvuloplastie par une approche transnasale orthograde sous guidage endoscopique à partir d'une vue nasopharyngée rétrofléchie. Pour éviter une nouvelle sténose, une sonde de Foley a été placée comme stent transitoire pendant 6 jours. Le chat s'est rétabli sans incident et était asymptomatique après le retrait du stent. Le réexamen endoscopique effectué 5 mois plus tard a confirmé la persistance de l'ouverture et de la perméabilité de la voie choanale droite générée. Le chat reste asymptomatique 10 mois après l'intervention. La dilatation endoscopique transnasale par ballonnet et la pose d'une endoprothèse transitoire dans le cas d'une atrésie des choanes est une procédure peu invasive et relativement simple dont le succès peut être durable.

Mots clés: chat, empyème sous-dural, valvuloplastie, sténose nasopharyngée, sonde de Foley

Dilatazione con palloncino e stent transitorio di un'atresia coanale membranosa unilaterale in un gatto British Shorthair affetto da rinite purulenta cronica e meningoencefalite ascendente

L'atresia coanale è una rara anomalia congenita negli esseri umani e negli animali, caratterizzata dall'assenza di comunicazione di una o entrambe le cavità nasali con la rinofaringe. La gravità dei segni clinici dipende dalla presenza di stenosi unilaterale o bilaterale, nonché dalle comorbidità. Con l'atresia bilaterale, la respirazione può essere gravemente compromessa, specialmente durante il sonno, poiché il flusso d'aria può avvenire solo respirando attraverso la bocca aperta. Sono state descritte varie modalità terapeutiche nelle persone e adattate agli animali. Tutti i trattamenti possono essere associati a complicazioni, la più importante delle quali è la formazione di cicatrici post-terapeutiche con nuove stenosi. Questo studio descrive un gatto British Shorthair di 10 mesi con scarico nasale seroso unilaterale cronico che è diventato mucopurulento. Quando si sono sviluppati segni neurologici acuti, il gatto è stato trasportato all'ospedale veterinario. Una diagnosi di atresia coanale primaria e membranosa del lato destro è stata raggiunta tramite tomografia computerizzata (TC) e rinofaringoscopia (posteriore). Le modifiche secondarie includono una rinite distruttiva con progressione al sistema nervoso centrale con un empiema subdurale e una meningoencefalite. Sono state sospettate modifiche retiniche e broncopolmonite da aspirazione come complicazioni aggiuntive. Dopo il recupero dalle infezioni secondarie, l'ostruzione membranosa è stata perforata e dilatata utilizzando un palloncino valvoloplastico con un approccio transnasale ortogonale sotto guida endoscopica da una vista retroflessa della rinofaringe. Per prevenire una nuova stenosi, è stato posizionato un catetere Foley come stent temporaneo per 6 giorni. Il gatto si è ripreso senza problemi ed è rimasto asintomatico dopo la rimozione del catetere. Un seguente esame endoscopico dopo 5 mesi ha confermato la persistenza dell'apertura e la pervietà del passaggio coanale destro generato. Il gatto è rimasto asintomatico 10 mesi dopo l'intervento. La dilatazione endoscopica con palloncino transnasale e il posizionamento temporaneo di uno stent per l'atresia coanale è una procedura minimamente invasiva e relativamente semplice con un successo tendenzialmente duraturo.

Parole chiave: gatto, empiema subdurale, valvuloplastica, stenosi rinofaringea, catetere di Foley

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