Surgical correction of a bilateral congenital nasal pyriform aperture stenosis in a French Bulldog

Chirurgische correctie van bilaterale congenitale stenose van de nasale pyriforme opening bij een Franse buldog

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ABSTRACT

A four-month-old, male, intact French Bulldog was presented with bilateral purulent nasal discharge and difficulties in breathing. A congenital nasal pyriform aperture stenosis with closure of both nasal cavities was diagnosed. The aperture was surgically opened through a midline approach, and a covered metallic stent (CMS) was inserted in each nasal cavity to prevent the newly constructed airway from obstructing. Seven weeks after implantation, the left nasal cavity was obstructed by granulation tissue. Further diagnostics revealed that the left stent had migrated caudally. The aperture had to be reopened and a new stent was placed. The right stent was removed endoscopically at that time, the left stent was removed six weeks after implantation. Four weeks after the removal of the left stent, the dog had normal airflow through both nares with very mild serous nasal discharge.

SAMENVATTING

Een vier maanden oude, mannelijke Franse buldog met klachten van bilaterale purulente neusvloei werd doorverwezen voor verder onderzoek. Bilaterale congenitale stenose van de apertura pyriforemis werd gediagnosticeerd en chirurgisch gecorrigeerd. Vervolgens werd een bedekte metaalstent (“covered metallic stent“, CMS) in iedere neusopening geplaatst om herstenose te voorkomen. Zeven weken na de ingreep werd er recidief van de stenose vastgesteld ter hoogte van de linker neusopening door granulatieweefsel. Verder onderzoek toonde aan dat de linker stent naar caudaal gemigreerd was. De linker apertura werd opnieuw chirurgisch geopend en een nieuwe stent werd geplaatst. Tijdens deze ingreep werd de rechter stent endoscopisch verwijderd. De linker stent werd zes weken later eveneens verwijderd. Vier weken na het verwijderen van de linker stent kon de patiënt door beide neusgaten ademen en vertoonde milde sereuze neusvloei.

INTRODUCTION

Craniofacial development is a well-coordinated and highly complex process. In early embryonic development, there are seven prominences forming the face. The forehead, the philtrum of the upper lip, the primary palate and the middle of the nose are produced by the frontonasal prominence whereas the sides of the nose are formed by the lateral nasal and the lower jaw by the mandibular prominences. The maxillary prominences form the lips, secondary palate and the sides of the face (Tapadia et al., 2005). Disturbances of this complex process can induce a variety of abnormalities (Nemec et al., 2015). Localized congenital abnormalities of the nasal cavity are rare, and as in other organs, it is difficult to determine the actual cause (Lopez, 2006). In this case report, a malformation is described, known in human medicine as congenital nasal pyriform aperture stenosis (CNPAS). CNPAS is a rare cause of neonatal obstruction leading to respiratory distress (Moreddu et al., 2016). Both nasal cavities were surgically opened and covered metallic stents (CMS) were placed in each cavum to prevent the newly constructed airway from recurrent stenosis. To the authors’ knowledge this has not been previously described.

CASE DESCRIPTION

A four-month-old, male, intact French Bulldog was presented with a six-week history of bilateral purulent nasal discharge and continuous open-mouth breathing. The owner mentioned the dog needed a thumb or a toy in his mouth before being able to fall asleep. Additionally, he displayed episodes of regurgitation. At the time of presentation, he was clearly smaller than his litter mates. Previous bacteriologic (aerobic and anaerobic) as well as fungal cultures had been performed by the referring veterinarian after which the patient was treated with an appropriate antimicrobial drug (unknown product and dosage). The nasal discharge initially improved with antimicrobial drug therapy; however recurred multiple times after discontinuing the therapy. No further diagnostic tests were performed by the referring veterinarian. On initial physical examination, the dog had a breed specific shortened skull and muzzle, with a craniofacial ratio of 0.24. His body condition score was slightly reduced (3 out of 9). An absence of airflow through both nares resulted in open-mouth breathing at a normal frequency (20/min). A laryngeal and pharyngeal stridor and bilateral purulent nasal discharge were also present. Due to the absence of airflow, a stertor was not present. The remainder of the physical examination was unremarkable. A complete blood count and serum biochemistry profile were within normal ranges. The dog was anesthetized for diagnostic imaging. Induction was initiated with an intravenous bolus of propofol (4 mg/kg, PropoVet Multidose, Ecuphar GmbH, Greifswald, Germany), followed by diazepam (0.5 mg/kg, Diazepam, Merckle, Ulm, Germany) and maintained with isoflurane (2%, Isoflo, Ecuphar GmbH, Greifswald, Germany) vaporized in oxygen.

Computed tomographic (CT) imaging was done using a 16-slice spiral CT scanner (Toshiba). The dog

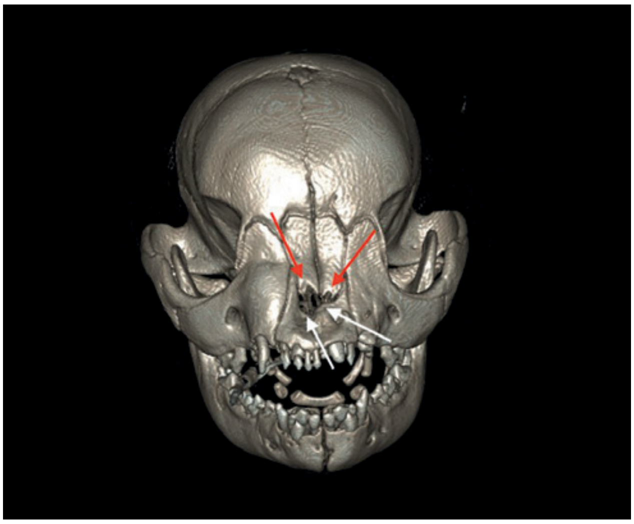

Figure 1. Three-dimensional, reconstructed CT image of the skull showing bilateral deformation of the nasal bone (red arrows) and incisive bone (white arrows).

was positioned in sternal recumbency. One-millimeter thick contiguous transverse CT skull images were obtained at 120 kV and 150 mA. The display field of view (DFOV) was 16.2 cm x 16.2 cm with a 512 × 512-pixel matrix with and without intravenous contrast (2 ml/kg, Accupaque 300, GE Healthcare Buchler GmbH & Co.KG, Braunschweig, Germany). The images were analyzed in a soft tissue window (WW 350 and WL 100) and in a bone window (WW 3500 and WL 1000). Subsequently, a multiplanar reconstructive image was created.

A severe brachycephalic deformation of the rostral cranium with a high-grade shortening of the left maxilla was shown on the CT reconstruction images. As a result of this shortening, a deformation with an oblique orientation of the incisive bone and the nasal bone was present (Figure 1). Both nasal and incisive

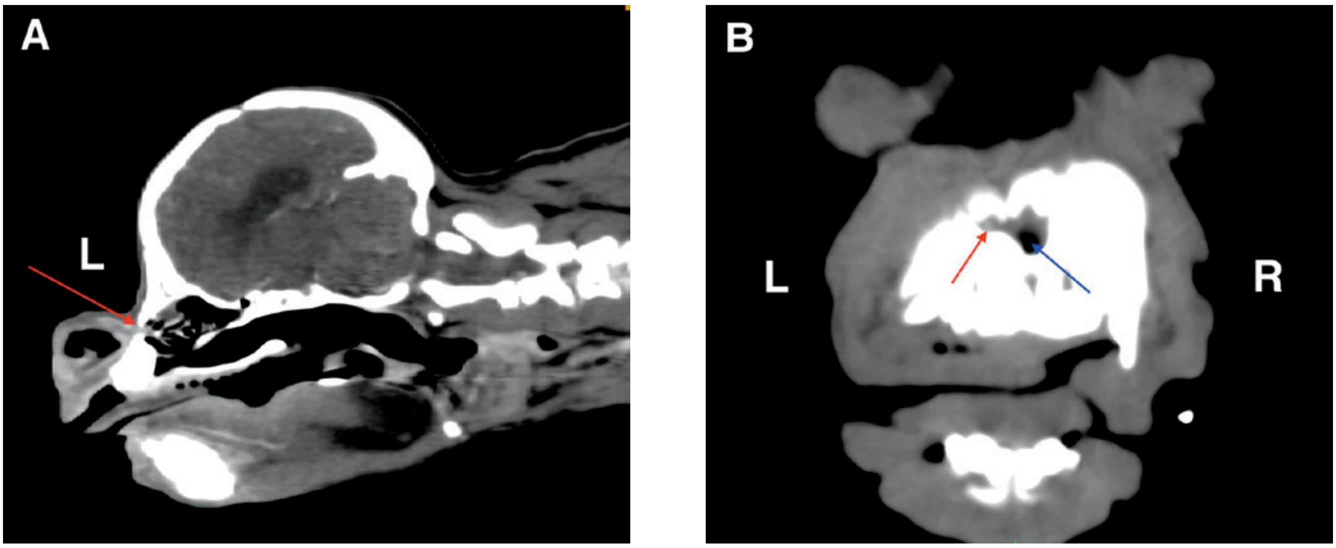

Figure 2. A. Sagittal multiplane reconstructed CT image through the left nasal cavity showing the stenotic pyriform aperture (red arrow). B. Transverse multiplane reconstructed CT image through both nasal cavities. The completely obstructed left pyriform aperture (red arrow) and the 2 mm opening of the right pyriform aperture (blue arrow) are shown.

bones had a concave shape and narrowed the nasal cavity up to a few millimeters. Additionally, both nasal cavities were narrowed further by a dense soft tissue material, closing the right nasal cavity up to 2 mm and the left cavum nasi completely. (Figures 2A and 2B). The nasal planum was developed normally. The frontal sinuses were absent on both sides and the endoturbinates showed retrograde growth into the nasofarynx.

For surgical correction, the dog was premedicated with methadon (0.5 mg/kg, Comfortan, Eurovet Animal Health B.V., the Netherlands) induced with intravenous propofol (4 mg/kg, PropoVet Multidose, EcupharGmbH, Greifswald, Germany) and anesthesia was maintained with isoflurane (2%, Isoflo, EcupharGmbH, Greifswald, Germany) vaporized in oxygen. Additionally, a constant rate infusion with fentanyl (8 µg/kg/h, Fentadon, Eurovet Animal Health B.V., the Netherlands) was administered during surgery. The procedure was performed by the first author. The dog was positioned in sternal recumbency. A midline skin incision over the nasal bone was performed. The incision was spread using Gelpi retractors. The dorsal and lateral nasal ligaments were sharply dissected and the nasal planum was dissected bluntly from the bone (Figure 3). The right opening of the cavum nasi was enlarged by removing excessive fibrous soft tissue and incisive bone as well as nasal bone. The nasal planum cartilage was macroscopically normal. The stenotic left nasal cavity was opened using a burr, and was consecutively enlarged as described above (Figures 4 and 5). The surgical field was rinsed with lukewarm, isotonic saline (NaCl 0.9%, B. Braun, Melsungen, Germany), and the incision was closed routinely. Based on preoperative CT measurements of the nasal cavity, a covered metallic stent (Conovet, Achern, Germany) of 10 mm diameter (20% larger than the estimated 8-mm end result) was placed in each cavity (Figures 6A and 6B) and correct stent placement for all three dimensions was controlled by using computed tomographic imaging (Figures 7). No intra-operative complications occurred.

The day after surgery, the dog had airflow through both nares and showed no signs of open-mouth breathing anymore. Amoxicillin-clavulanic acid (8.75 mg/kg, Synulox; Zoetis Deutschland GmbH, Berlin, Germany) and meloxicam (0.2 mg/kg, Metacam, Boehringer Ingelheim Vetmedica GmbH, Ingelheim/Rhein, Germany) were administered subcutaneously. The second day postoperatively, the dog was discharged and oral medication was continued with amoxicillin-clavulanic acid (12.5 mg/kg, twice daily, Synulox, Zoetis Deutschland GmbH, Berlin, Germany) and meloxicam (0.1 mg/kg, Metacam, Boehringer Ingelheim Vetmedica GmbH, Ingelheim/Rhein, Germany) for ten days.

Two weeks after surgical correction, the dog was reassessed in the clinic. On physical examination, he was alert and active with vital signs within nor-

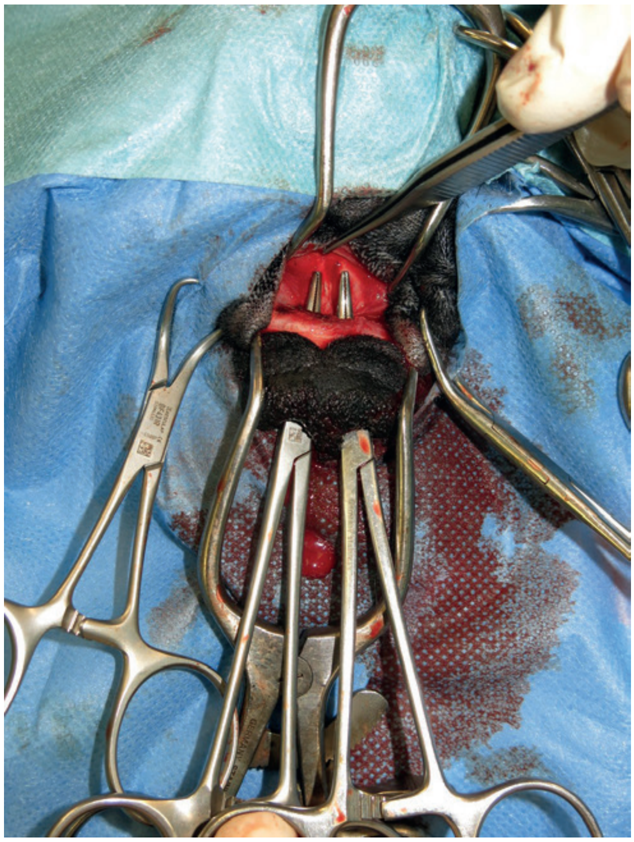

Figure 3. Intraoperative view of the dorsal midline incision. The incision was spread using Gelpi retractors, showing cranial and caudal V-shaped endings. The nasal planum was dissected bluntly from the nasal bone.

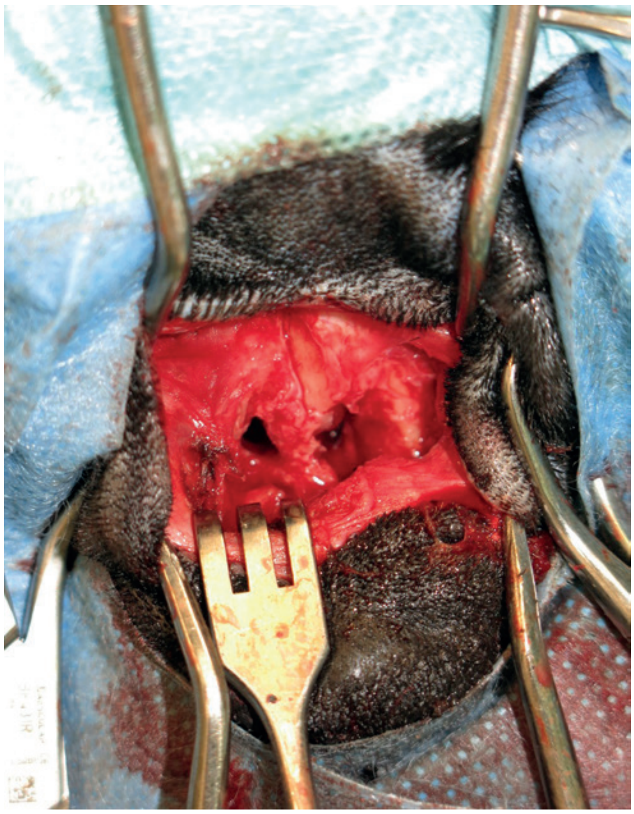

Figure 4. The stenotic nasal cavities after opening, using a burr and removing redundant soft tissue and bone.

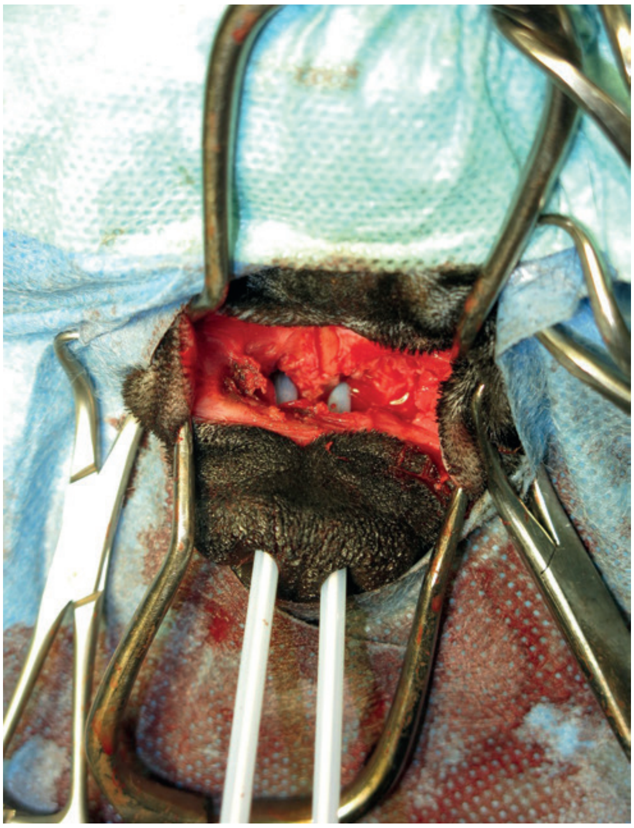

Figure 5. Assessment of the correct diameter of the newly created openings before final placement of the metallic stents.

mal ranges. He was breathing through both nares but had bilateral purulent discharge. Treatment with oral amoxicillin-clavulanic acid (12.5 mg/kg twice daily, Synulox, Zoetis Deutschland GmbH, Berlin, Germany) was restarted and continued for seven days. This resolved the purulent discharge initially but on recheck examination seven weeks later, a purulent discharge on the right side recurred and absence of nasal airflow on the left side was detected. The dog was examined under anesthesia using video endoscopy. The same anesthesia protocol of the previous CT examination was used. Endoscopic examination of the right nasal opening showed the stent in situ with a high degree of purulent discharge. The cavity was flushed with isotonic saline (NaCl 0.9%, B. Braun, Melsungen, Germany). The left cavum nasi was completely closed by granulation tissue at the level of the rostral tip of the nasal bone (rostral process), which was not visible during clinical examination. The subsequent CT showed a caudal migration of the left stent and recurrent stenosis of the left nasal cavity.

The left nasal cavity was reopened using the same technique as before. The migrated stent was removed and a new stent of the same dimension was inserted. The right stent was removed. The dog made an uneventful recovery. The newly placed left stent was removed six weeks after implantation.

Five months after implantation of the first stent and four weeks after removing of the left stent, the dog was bright, alert and normal airflow through both nares with very mild serous nasal discharge was detected on physical examination. His reduced body condition had improved (5 out of 9) and the owner reported very rare episodes of regurgitation after surgery. Furthermore, during walks and during sleeping, the dog could breathe through his nose.

DISCUSSION

In the case presented here, a rare congenital abnormality of the nose was diagnosed in a four-month-old French Bulldog. Intranasal anomalies leading to anatomical obstruction of the intranasal airways in brachycephalic breeds are well described (Oechtering et al., 2016). This malformation called CNPAS, clinically first described in 1989 in human medicine, has not been described in dogs yet.

Craniofacial formation is a very complex and exact concerted process. Impairment of this developmental process can induce a variety of abnormalities (Nemec et al., 2015). In humans, this rare condition occurs in newborns and is caused by a premature fusion and overgrowth of the nasal medial processes of the maxilla; however, the exact etiology remains unclear (Brown et al., 1989; Belden et al. 1999). Depending on the severity of the patient's symptoms conservative treatment can be attempted. Nevertheless, in most cases, surgery is required (Moreddu et al., 2016). Since there was a complete closure of the aperture in this French Bulldog, surgery was required and the goal of treatment was to allow nasal breathing, to reduce the upper airway resistance and consequently to increase quality of life. To enable sufficient nasal breathing, the nasal bone was opened using a burr, and subsequently the obstructing soft tissue was removed. The primary goal after surgical opening of the nose was to prevent recurrent stenosis due to stricture formation or granulation tissue. Surgical treatment for humans with CNPAS is classically followed by nasal stenting in order to maintain patency and prevent adhesions of the newly established airway (Chander et al., 2015; Moreddu et al., 2016). For intraluminal stent placement in the trachea, a 10%-20% greater stent diameter than the maximum diameter is recommended to prevent stent migration (Beal, 2013). Therefore, a stent 20% larger than the estimated diameter of the new nasal openings was chosen in this clinical case. Despite this larger diameter, the left nasal stent migrated caudally, granulation tissue closed the surgical opening again and surgical intervention was required. The strong negative inspiratory breathing pressure of brachycephalic dogs might have facilitated the caudal migration of the stent. The left nasal cavity had to be reopened, the stent removed and a new one inserted. Retrospectively, the new bony opening of the left nasal cavity was not enlarged adequately. Therefore,

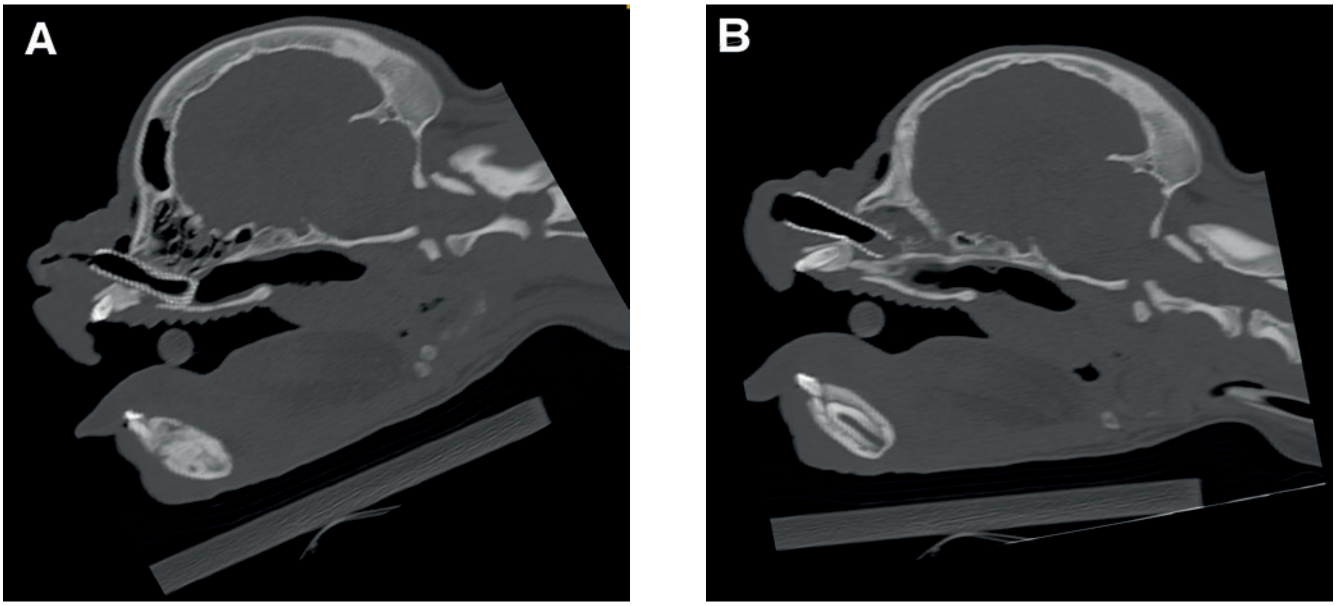

Figure 6. Sagittal reconstructed CT image after placement of both metallic stents into the nasal cavity. A. The right nasal stent. B. The left stent in situ.

the original stent could not expand sufficiently and subsequently did not come into full contact with the caudally located mucosa in the somewhat larger diameter nasal cavity, which can cause infection. During the second procedure, the newly created opening was slightly larger. The same diameter stent could therefore expand more than previously, made better contact with the caudal nasal mucosa, and consequently, might have prevented stent migration.

For the treatment of nasopharyngeal stenosis and imperforate nasopharynx, placement of uncovered (UMS) and covered metallic nasal stents have been described. CMS seem to prevent tissue ingrowth, bending and stent fracture, but unlike UMS, they are not incorporated in the mucosa and therefore, foreign body reaction and infection are more likely to occur (Burdick et al., 2018). Even in humans, given the rarity of the disease, no standard for the size and duration of post-surgical stenting period has been described, but nasal stents are generally left in place for two to four weeks (Moreddu et al., 2016). In this case, CMS were chosen to prevent abundant granulation tissue ingrowth and recurrent stenosis of the nasal cavity.

Although pathogenic infections are usually not increased after correct tracheal stent placement (Lesnikowski et al., 2020), an infection might have caused problems after intraluminal nasal stenting in this patient. The purulent discharge occurred on both sides after stent placement. The lack of contact between stent and mucosa leads to accumulation of mucus, fluid, tissue and subsequent infection. In tracheal stenting, bacterial infection has been associated with a decreased mucosal integration thus resulting in coughing or stent migration (Durant et al., 2012). The lack of contact between the stent and nasal mucosa in this case might have facilitated stent migration in the left cavum nasi. In tracheal stenting, inflammatory tissue formation that may or may not be associated with bacterial tracheitis has been a documented problem in some dogs undergoing stent placement. Some animals with this complication may respond to antimicrobial treatment followed by corticosteroid therapy (Beal, 2013). In the dog of the present case, purulent nasal discharge improved initially with antimicrobial treatment but resolved only after stent removal. Corticosteroid therapy was not considered in this patient. Since there is only limited benefit of performing cytology bacterial culture of nasal discharges to diagnose bacterial infection and guide the antimicrobial

Figure 7. Three-dimensional reconstructed CT image (front view) after placement of both metallic stents.

choice, no further bacteriological examination was performed and a therapy with amoxicillin-clavulanate as a first-line antimicrobial treatment option was chosen (Lappin et al., 2017). Retrospectively, in the view of antimicrobial resistance, a local lavage might have been sufficient.

The concurrent prevalence of gastrointestinal tract problems accompanied by upper respiratory disease in brachycephalic breeds is well established (Poncet et al., 2005). The initial examination of this dog showed him to have retarded growth and a slightly reduced body condition with frequent episodes of regurgitation. The absence of nasal air passage resulted in highly increased negative lower airway pressure and therefore worsened his gastrointestinal signs. (Freiche et al., 2021; Poncet et al., 2006). After surgical treatment of the nasal airway passage, the gastrointestinal clinical signs resolved, and subsequently his body condition improved.

CONCLUSION

Despite stent migration in this case, placement of a CMS prevented the nasal cavity from closing and patency was maintained after corrective surgery. Further cases are necessary to evaluate the use of these stents to treat similar malformations.

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