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CASE REPORT

Companion or pet animals

Diagnosis and surgical management of a type IV*b* dermoid sinus in a Cavalier King Charles Spaniel

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Abstract

A 6-year-old, male, neutered Cavalier King Charles Spaniel dog was referred with a 4-year history of an intermittently draining fistula on the midline of the frontal bones of the skull. Magnetic resonance imaging and computed tomography scan of the head demonstrated an extra-axial mass in the midline between the frontal lobes, contacting the meninges and extending rostrally through a defect in the frontal bones. The mass was surgically excised via a frontal craniotomy. Histopathology of the mass demonstrated stratified squamous and keratinising epithelium with adnexal structures, consistent with the diagnosis of a dermoid sinus. Previous reports have documented dermoid sinuses in the nasal region in dogs and cats with intracranial extension. This case report documents a dermoid sinus in the frontal region with intracranial extension in a dog, confirmed by advanced diagnostic imaging, histopathology, and successfully treated surgically.

KEYWORDS

dermatology, neuroimaging, neurology, neuropathology, neurosurgery

BACKGROUND

A dermoid sinus is a congenital tubular skin defect resulting from incomplete separation of the skin from the neural tube during embryological development.[1] Dermoid sinuses are well described in the Rhodesian Ridgeback breed[2] in which an autosomal dominant mutation causes the dorsal ridge and predisposes the breed to dermoid sinus formation.[3] Dermoid sinus formation has been reported in other breeds in the nasal,[4,5] cervical,[6] thoracic,[7–10] lumbosacral[11] and sacro-coccygeal[12] regions, but rarely in the head region.[13–16] Previous case reports described dermoid sinuses in the frontal, parietal, parieto-occipital and fronto-occipital regions in dogs.[13–16] Only two case reports documented a dermoid sinus in the nasal region with intracranial extension in a dog and cat.[4,17] This case report describes the advanced imaging, histopathological diagnosis and successful surgical management of a dermoid sinus contacting the meninges and extending rostrally through a defect in the frontal bones, in a Cavalier King Charles Spaniel dog. Based on previously proposed classifications, this is consistent with a type IV*b* dermoid sinus.[13,18]

CASE PRESENTATION

A 6-year-old, male, neutered Cavalier King Charles Spaniel dog was referred with a 4-year history of an intermittently exudative fistula on the midline of the frontal bone of the skull. The lesion initially appeared as a soft tissue swelling with serous discharge. The referring veterinarian had surgically excised the exudative fistula multiple times. Following recurrence of the fistula, the referring veterinarian performed a computed tomography (CT) scan of the head. The CT demonstrated a bony defect between the frontal bone and a sinus tract extending from the skin to the cribriform plate. A dermoid sinus was suspected; however, further surgical management was not pursued at that time due to owner unwillingness. One year later, the exudative fistula recurred, and referral for further investigations and treatment was pursued.

INVESTIGATIONS

Physical examination demonstrated a 3 mm fistula on the midline of the frontal bones of the skull (Figure 1a). A mild serous discharge was evident. The remainder of the physical examination was within normal limits. Neurological examination was unremarkable.

Haematology demonstrated thrombocytopenia (platelet count 138 × 10^9/L; reference interval: 200–500 × 10^9/L). Examination of a fresh blood smear demonstrated adequate platelet number. Biochemistry demonstrated an elevated alkaline phosphatase (ALKP 178 U/L; reference interval: 12–83 U/L). The remainder of the haematology and biochemistry

was within normal range. No analysis of the discharge was performed due to reduced volume.

A magnetic resonance imaging (MRI) study of the head was performed with a 1.5 Tesla MRI machine (Signa, General Electric) while under general anaesthesia. The dog was premedicated with butorphanol (0.3 mg/kg; Dolorex, MSD) intravenously (IV). General anaesthesia was induced with midazolam (0.2 mg/kg; Midazolam, Hameln) and propofol (1 mg/kg; PropoFlo, Abbott) IV to effect, and maintained on 2% isoflurane with 100% oxygen. The MRI study included T2W sagittal, T2W transverse, fluid-attenuated inversion recovery (FLAIR) transverse, T2*GE transverse, short tau inversion recovery (STIR) transverse, as well as T1W pre- and post-contrast sagittal, transverse and dorsal sequences (Figure 2a–k).

The MRI study showed the presence of an extra-axial mass in the midline between the frontal lobes, in contact with the falx cerebri with extension rostrally through a frontal bone defect into the left frontal sinus and then reaching the skin surface. The mass was mildly compressing the adjacent brain tissue at the level of the falx cerebri displacing both frontal lobes laterally and caudally. The mass appeared iso-/irregularly hyperintense to grey matter in T2W and STIR sequences, heterogeneously T1W and FLAIR iso- to hypointense, and heterogeneously hyperintense in the T2*GE sequence, with a thin peripheral rim characterised by 'signal void artefact'. In the T1W post-contrast sequences, the mass and its rostral extension through the left frontal sinus and subcutaneous tissue showed mild peripheral contrast enhancement delineating a thin capsule, while its central aspect did not show any contrast enhancement. The perilesional subcutaneous tissue, extending rostrally to the skin surface, was also diffusely contrast-enhancing. In addition, there was contrast enhancement of the meninges of the falx cerebri and of frontal lobes in contact with the extra-axial mass. The MRI study also revealed that the vermis of the cerebellum was impacted into the foramen magnum and partially herniated through it. Additional MRI findings included the presence of a mild flattening of the supraoccipital bone, a dorsal atlantoaxial band, marked dilation of the central canal of the cranial cervical spinal cord (>2 mm of maximum height) and the presence of T1W and T2W hyperintense and non-contrast-enhancing material in the tympanic bulla bilaterally. The MRI findings were compatible with a suspected dermoid sinus with intracranial extension or neoplastic transformation of a dermoid cyst, as well as caudal occipital malformation syndrome, cervical syringomyelia and suspected bilateral secretory otitis media.

Following the MRI study, under the same general anaesthesia and with the patient positioned in sternal recumbency, a CT scan (16-slice multidetector, helical GE CT) of the head with bone and soft-tissue algorithms with 1.25 mm slice thickness was performed for surgical planning. The CT demonstrated a midline fusion defect between the frontal bones extending through the cribriform plate, allowing communication with the subcutaneous tissues (Figure 3a–c).

Cisternal cerebrospinal fluid (CSF) was collected. The CSF analysis demonstrated a clear and colourless fluid. An elevated total nucleated cell count (8 cells/μL; reference: <5 cells/μL) and protein (39.2 mg/dL; reference: <25 mg/dL) were demonstrated. Cytology revealed a mononuclear pleocytosis. Bacterial culture demonstrated no growth.

LEARNING POINTS/TAKE-HOME MESSAGES

- Type IV*b* dermoid sinus can be found in the frontal bone region in dogs.
- The presence of a draining fistula at the level of the head should raise suspicion of a dermoid sinus.
- Computed tomography and magnetic resonance imaging scans of the head can facilitate further characterisation of the sinus extent.

DIFFERENTIAL DIAGNOSIS

Based on the clinical signs and diagnostic imaging findings, a dermoid sinus or a neoplastic transformation of a dermoid cyst was considered the most likely diagnosis. Differential diagnoses including foreign body or abscessation were considered less likely due to the chronicity of clinical signs.

TREATMENT

Surgical excision of the suspected dermoid sinus was elected by the owner to reach a definitive diagnosis and to prevent the recurrence of the fistula. General anaesthesia was induced with a combination of methadone (0.2 mg/kg IV; Comfortan, Dechra), midazolam (0.2 mg/kg IV; Midazolam, Hameln) and alfaxalone (2 mg/kg IV; Alfaxan, Jurox). General anaesthesia was maintained with 2.2% sevoflurane in 100% oxygen. Analgesia was provided with intraoperative continuous rate infusions of 5 µg/kg/h fentanyl (Fentadon, Dechra) and 10 µg/kg/min ketamine (Anesketin, Dechra). Antibiotic prophylaxis was achieved with cefuroxime (20 mg/kg IV; Zinacef, Glaxo Smith Kline) and administered every 90 minutes intraoperatively.

The patient was positioned in sternal recumbency with the head elevated to avoid jugular venous compression. Following routine surgical preparation, an elliptical incision was made around the discharging fistula, from the caudal aspect of the nasal bone to the caudal aspect of the frontal sinuses at the level of the bregma. The sinus tract was sharply and bluntly dissected free from the surrounding soft tissues and frontal sinuses. A 4 Fr Tom Cat urinary catheter facilitated orientation of the sinus tract. The sinus tract was retracted exposing the internal table of the frontal bone. An oval-shaped bone defect was identified in the internal table of the frontal bone, through which the sinus tract extended. The bony defect was enlarged with a pneumatic neurosurgical drill (Surgairtome Two Hall, Conmed) to facilitate excision of the remaining sinus tract, which was separated from the meninges of the falx cerebri at the level of the frontal lobes (Figure 1b–d). Before reaching the dura mater, 0.5 g/kg of 10% mannitol (Mannitol, Viaflo) was administered IV over 15 minutes.

During dissection of the sinus tract, the lining was ruptured, exposing hair, thick white exudate and debris within the frontal sinus. The surgical site was copiously lavaged with sterile saline (NaCl 0.9%, Braun), and a swab for bacterial culture/sensitivity was taken from the surgical site. A synthetic dural graft (Lyoplant, Braun) was placed over the exposed frontal lobes. The surgical incision was closed routinely. A

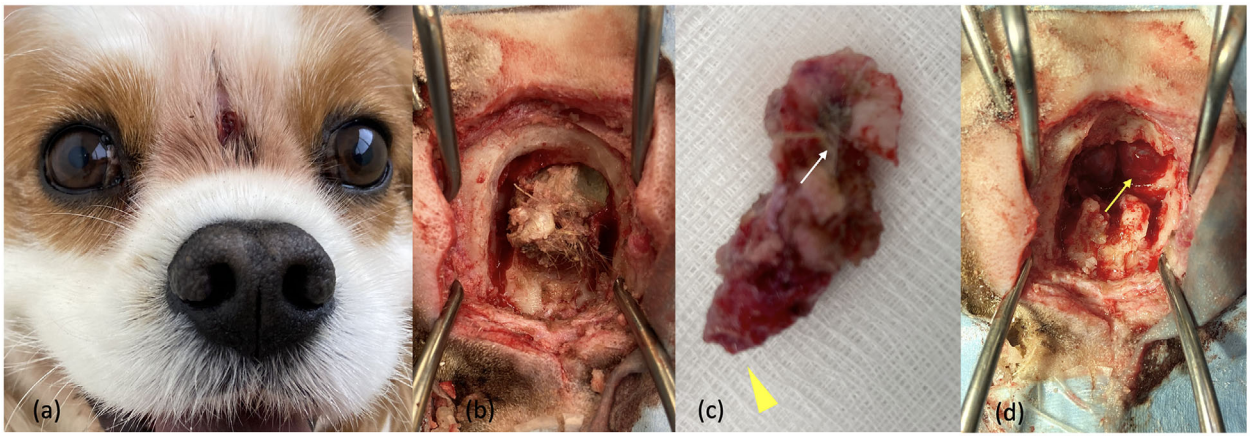

FIGURE 1 (a) Photograph demonstrating the draining fistula at the level of the frontal bones. (b) Intraoperative photograph of the dermoid sinus, displaying connective tissue, and hair. (c) Surgically excised dermoid sinus, showing the rostral aspect of the fistula at the skin surface (white arrow), and the caudal aspect of the mass (yellow arrowhead), which was in contact with the meninges of the falx cerebri. (d) Intraoperative photograph of the trans-frontal skull defect displaying the meninges of the falx cerebri (yellow arrow) following the surgical excision.

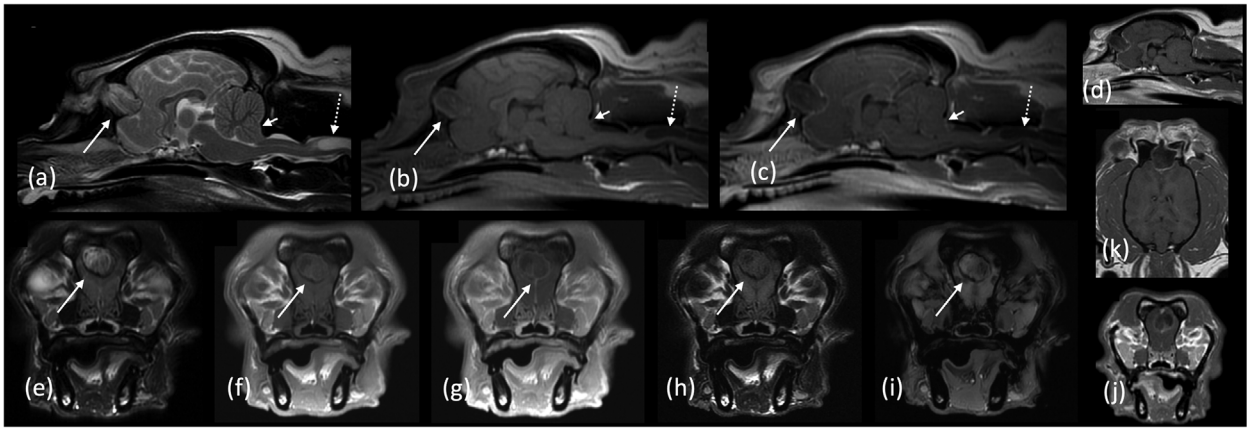

FIGURE 2 The (a) T2W, (b) T1W pre-contrast and (c) T1W post-contrast, (d) T1 three-dimensional cube sagittal multiplanar reconstruction magnetic resonance imaging (MRI) images, and (e) T2W, (f) T1W pre-contrast and (g) post-contrast T1W, (h) fluid-attenuated inversion recovery, (i) T2*GE, (j) multiplanar reconstruction in transverse and (k) dorsal multiplanar reconstruction MRI images showing the dermoid sinus (long white arrows) in the midline of the frontal bones, being in contact with the meninges of the falx cerebri and extending rostrally through a frontal bone defect into the left frontal sinus and then reaching the skin surface. Caudal occipital malformation syndrome (short white arrows) and cervical syringomyelia (dotted white arrows) in (a), (b) and (c).

post-operative CT scan was performed, confirming complete excision of the sinus tract.

The excised tissue was submitted for histopathology, which confirmed keratinising stratified squamous epithelium and associated adnexal structures (Figure 4a,b). The epithelial lining was extensively ulcerated, and replaced by fibrovascular connective tissue with acute haemorrhage. There was an extension of maturing granulation tissue infiltrated by abundant mixed lymphocytes, plasma cells, neutrophils and macrophages. These findings were compatible with the diagnosis of a dermoid sinus.

OUTCOME AND FOLLOW-UP

The dog recovered uneventfully from surgery. Post-operative analgesia was provided with paracetamol (10 mg/kg IV; Paracetamol, Braun) at 12-hour intervals, as well as methadone (0.2 mg/kg IV; Comfortan, Dechra) at 4-hour intervals; the latter reduced 12 hours post-operatively to 0.1 mg/kg at 4-hour intervals for a further 24 hours and was then discontinued. Amoxicillin/clavulanic acid (20 mg/kg orally; Clavaseptin, Vetoqinol) was administered at 12-hour intervals until receipt of a negative intraoperative bacterial culture. The dog was discharged 3 days following the surgery. A 4-week post-operative check confirmed complete healing of the surgical site.

Five months following surgical excision of the dermoid sinus, a 3-day history of a mild serous discharge at the surgical site was reported. The surgical scar was mildly erythematous. Palpation demonstrated no pain or swelling. Differential diagnoses included cellulitis, suture reaction, surgical site infection or incomplete excision of the sinus tract. An MRI of the head was repeated, which demonstrated no recurrence of the dermoid sinus (Figure 5a–c). The cause of the serous discharge was unknown; however, resolved with no treatment within 3 days. At the time of writing this report, a year later, there has been no recurrence of the serous discharge.

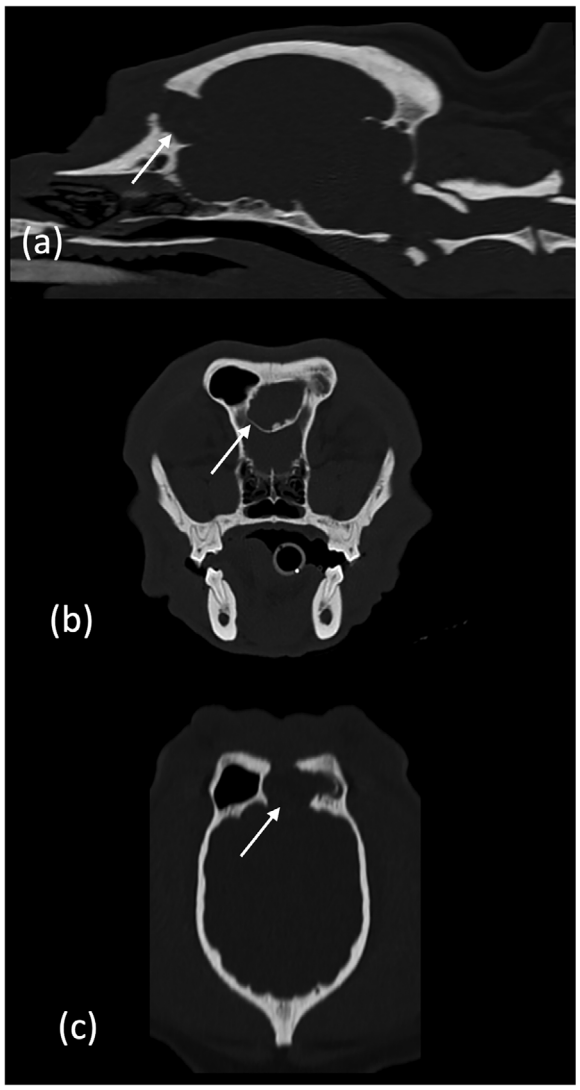

FIGURE 3 Computed tomography multiplanar reconstruction, (a) sagittal multiplanar reconstruction, (b) transverse multiplanar reconstruction, (c) dorsal multiplanar reconstruction, showing the mass extending from the frontal lobes towards the left frontal sinus through the internal table defect of the frontal bone.

DISCUSSION

This case report describes a dermoid sinus of the frontal bones with intracranial extension in a Cavalier King Charles Spaniel dog. To the best of the authors' knowledge, this has not been reported previously in dogs.

A classification of dermoid sinuses of the vertebral column in dogs has been proposed, with four types based on the degree of penetration into the subcutaneous tissues. Type I dermoid sinus terminates on the supraspinous ligament or nuchal ligament, type II consists of a superficial sac connected to the supraspinous ligament by a fibrous band, type III reaches the subcutaneous tissue, and type IV extends into the vertebral canal and is attached to the dura mater.[18] Furthermore, a distinct type of dermoid sinus, consisting of a closed epithelial-lined sac, was described as a true dermoid cyst or type V dermoid sinus.[7] A type VI dermoid sinus has been recently described, characterised by an open sinus tract extending to the supraspinous ligament and attached via a fibrous band to the dura mater.[9] Considering these classifications related to dermoid sinuses of the vertebral column only, Bornard et al.[13] proposed subtypes based on three anatomical locations. Dermoid sinuses located on the dorsal midline of the vertebral column were classified as subtype *a*, dermoid sinuses on the head (nose exempt) were classified as subtype *b* and dermoid sinuses in the nasal region were classified as subtype *c* (Figure 6).[13] Based on this classification, only four cases have previously been classified as subtype *b*.[13–16] Neither of these cases reported intracranial extension. One case reported a Rottweiler dog with a bilateral dermoid sinus in the parieto-occipital region.[13] A fibrous band connecting the subcutaneous tract to the periosteum bilaterally, at the level of the suture line between the parietal and occipital bones, was identified surgically.[13] Another case report documented multiple dermoid sinuses in the fronto-occipital region of a Saint Bernard dog.[14] This case had multiple sinus tracts with protruding hair.[14] The dermoid sinuses consisted of a combination of type V*b* and III*b*. The third case was a 5-month-old Australian shepherd dog that had a combination of follicular hamartomas with concurrent follicular cysts and dermoid sinuses in the parietal region, with the dermoid sinuses being classified as both III*b* and V*b*.[15] The fourth dog was a Miniature Schnauzer that had multiple dermoid sinuses in the frontal region surgically excised, and histopathologically confirmed to reach the subcutaneous tissue.[16] These cases reported no advanced imaging of the dermoid sinuses.[13–16] However, a recent case report documented a nasal dermoid sinus with intracranial extension with MRI in a cat.[17] Considering the proposed classification by Bornard et al.,[13] the cat would be classified as having a type IV*c* dermoid sinus.[17] Our case would be the first dog reported with a type IV*b* dermoid sinus.

The diagnosis of dermoid sinuses is based on clinical, advanced imaging and histopathological findings.[17] Depending on the location of the dermoid sinus, various neurological signs have been reported from pelvic limb neurological deficits to seizures.[11,17,19] In our reported case, there were no neurological signs. However, a draining fistula at the midline of the skull had been present for several years. The chronicity of the case is not surprising, as cases are usually initially referred for non-emergent dermatological complaints.[20] Clinically, a nasal pit, a grossly visible small opening has been described, and considered to be characteristic in humans with cranial midline dermoid sinuses.[21] Nasal pits refer to the remnants of primitive nares and nasal cavities found on the midline of the head.[21–23] This is explained by the incomplete separation of the epithelial cells from the neural crest during embryogenesis.[21,24] This hypothesis supports the location of dermoid sinuses found along the midline of the nose.[4,5,20] Although nasal location of dermoid sinuses has been reported in dogs,[4,5,20] considering the different dermoid sinus types, a sinus tract communicating with the skin is not always identifiable.[5] Anatomically, the skull is divided into several bones, originating from different group of cells from the mesoderm.[25] The bones of the skull develop further during the embryological phase and will fuse following birth. Theoretically, dermoid sinuses could occur at any fusion site, making the clinical diagnosis more challenging.[26,27] This theory was supported by previous veterinary cases, where sinus tracts were identified at the level of frontal, parietal, parieto-occipital and fronto-occipital regions[13–16]; however, more studies are warranted. Advanced imaging should be considered to confirm the presence, extent and distribution of the lesion.[11,17,19]

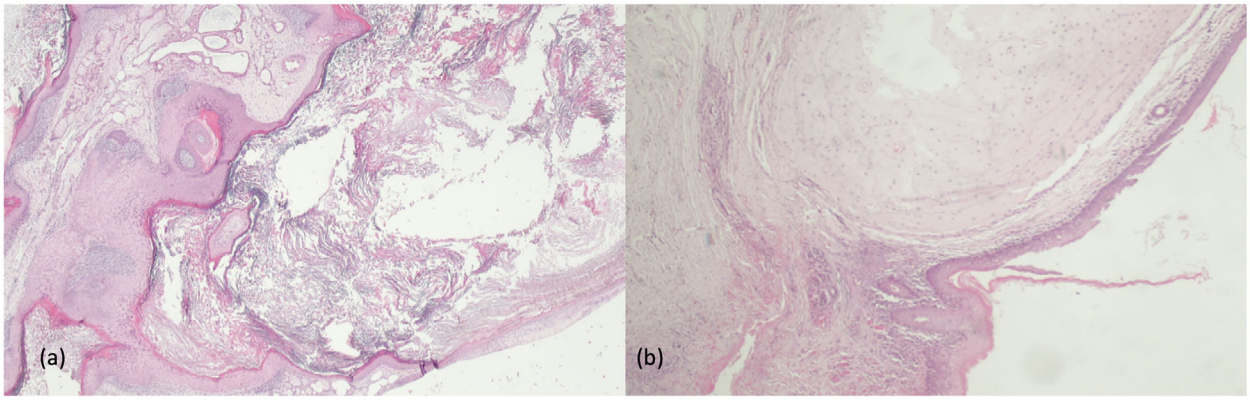

FIGURE 4 (a) Photomicrograph showing a partially cystic and irregular structure from the frontal sinus. The structure is formed of a stratified squamous epithelial lining and filled with lamellar fragments of keratin. There are peripheral adnexal structures including sebaceous glands, hair follicles and apocrine glands within the wall of the tissue (haematoxylin and eosin, 20×). (b) The stratified squamous and keratinizing epithelium are closely associated with and appear to extensively line woven bone (haematoxylin and eosin, 40×).

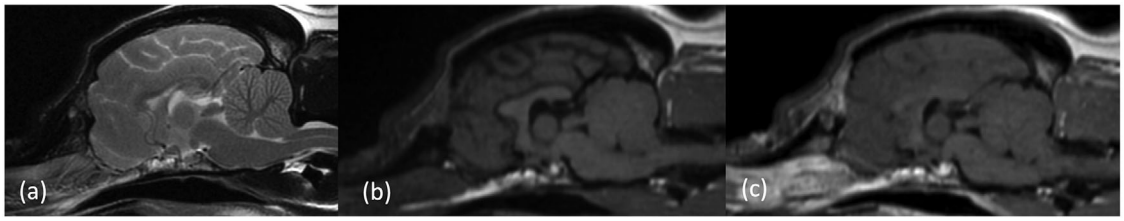

FIGURE 5 Post-surgical (a) T2W sagittal, (b) T1 three-dimensional (3D) cube pre-contrast sagittal multiplanar reconstruction, and (c) T1 3D cube post-contrast sagittal multiplanar reconstruction MRI images demonstrating no recurrence of the previous dermoid sinus.

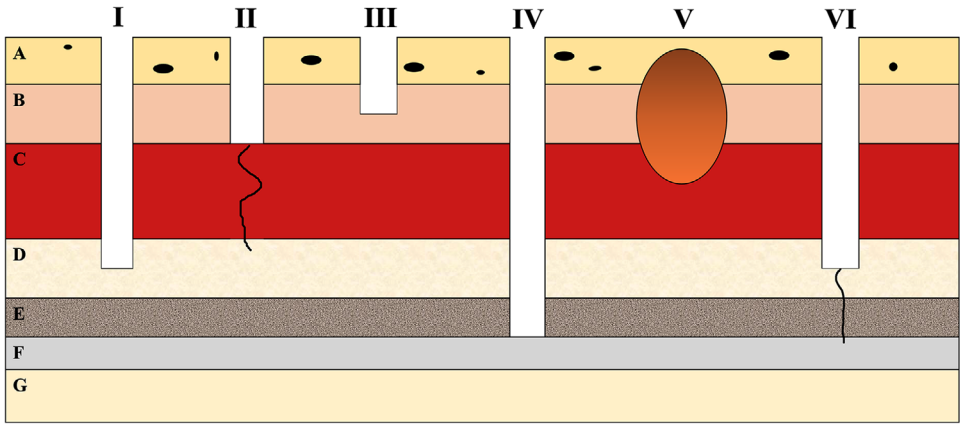

FIGURE 6 Dermoid sinus categories (modified following references[12,13,18] by the authors) based on the degree of penetration into the subcutaneous tissues. Type I dermoid sinus terminates on the supraspinous ligament; type II terminates on the supraspinous ligament via a fibrous band; type III reaches the subcutaneous tissue; type IV extends into the vertebral canal or into the cranial cavity; type V has no connection with the skin surface and has an epithelial-lined sac; and type VI penetrates until the supraspinous ligament and reaches the dura mater via a fibrous band. (a) Skin; (b) subcutaneous tissue; (c) muscle for subtype *a* and *b*; (d) supraspinous ligament for subtype *a* and cartilaginous nasal septum for subtype *c*; (e) periosteum of the cranial bones for subtype *b* and periosteum of vertebral column for subtype *a*; (f) dura mater; (g) brain for subtype *b* and *c*, spinal cord for subtype *a*.

Fistulograms, where contrast solution was infused via a catheter in the sinus tract, and lumbar myelograms were historically used for the diagnosis of dermoid sinuses.[9,13,24] Fistulograms were reported to be inaccurate in detecting the length of sinus tract, and could increase the risk of contaminating the spinal canal with sinus tract content.[9] Additionally, seizures were a reported side effect of lumbar myelograms.[28] However, advanced imaging is now commonly available, less invasive and can provide detailed information of the adjacent neurological, soft tissue and bony structures.[17] Due to the prolonged time since the initial CT scan, a repeat CT scan was performed to visualise the bony defect for surgical planning, while the MRI facilitated assessment of the underlying neural structures.

In our case, the dermoid sinus was T2W and STIR hyperintense, T1W and FLAIR iso- to hypointense, with contrast enhancement at the level of the connection with the frontal lobes on the MRI images. In the previous report documenting a cat with intracranial extension of the nasal dermoid sinus, heterogeneous hyperintensity on both T1W and T2W images

was observed on MRI of the head.[17] The MRI findings are similar to dermoid sinuses in the human literature, with T1W images signal intensities varying from hypo- to hyperintense, largely dependent on the fat content of the dermoid sinus.[29,30]

CSF analysis revealed a mild mononuclear pleocytosis, elevated protein concentration and a negative bacterial culture. CSF changes have not been previously reported in dogs diagnosed with dermoid sinuses in the head. Similar CSF findings have been reported in two dogs with dermoid sinuses of the vertebral column and in a dog with an intracranial epidermoid cyst.[11,19,31] Although bacterial culture of the CSF was negative in our case, due to the T1W post-contrast enhancement of the meninges of the falx cerebri, it remains possible that CSF inflammation was caused by secondary bacterial contamination from the type IV*b* dermoid sinus. Bacterial culture of the CSF was reported to be negative in a dog with a spinal dermoid sinus and inflammatory CSF.[19]

Epidermoid cysts can potentially cause chemical meningitis by the release of cholesterol breakdown products into the CSF.[32] This was reported in humans, as a rare recurrent aseptic meningitis, called Mollaret's meningitis. This is characterised by a polymorphonuclear or mononuclear pleocytosis in the CSF.[32] It is possible that similar pathophysiology induces the CSF inflammation of dogs with type IV dermoid sinuses. Although, this requires more thorough investigations, it is not uncommon in dogs to have negative bacterial culture, with more severe neurological inflammatory conditions, such as intracranial empyema[33] or bacterial meningitis.[34]

In the human literature, even in asymptomatic cases, aggressive treatment for complete resection of the dermoid sinus is advised to reduce chances of infection and development of neurological signs.[35] Based on the veterinary literature, surgical excision of a dermoid sinus, with the removal of the cystic lining, is considered curative with a good prognosis.[1,15,17] However, recurrence of the clinical signs can occur if excision is incomplete.[1] The development of a serous discharge 5 months post-operatively was a concern; however, repeat MRI demonstrated no evidence of remnants of the cystic lining or fluid accumulation. While the cause could not be confirmed, the serous discharge self-resolved without medical intervention. The cause remains unknown; however, there has been no further recurrence at the time of writing this report.

In conclusion, this paper is the first report of a type IV*b* dermoid sinus in the frontal region of a Cavalier King Charles Spaniel, confirmed by advanced imaging and histopathology, with successful surgical excision.

AUTHOR CONTRIBUTIONS

Cecilia-Gabriella Danciu, Chiara Briola, Daniella McCready and Sue Fitzmaurice conceived and designed the project. Cecilia-Gabriella Danciu, Chiara Briola, Veerle Volckaert, Rachel Pittaway, Daniella McCready and Sue Fitzmaurice acquired the data. Cecilia-Gabriella Danciu, Chiara Briola, Veerle Volckaert and Daniella McCready analysed and interpreted the data. Cecilia-Gabriella Danciu, Chiara Briola, Veerle Volckaert, Daniella McCready and Sue Fitzmaurice wrote the paper.

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CONFLICT OF INTEREST STATEMENT

The authors declare they have no conflicts of interest.

ETHICS STATEMENT

The authors confirm that the ethical policies of the journal, as noted on the journal's author guidelines page, have been adhered to. No ethical approval was required as this is a case report with no original research data. The owner gave informed written consent for all the investigations.

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MULTIPLE-CHOICE QUESTION

What is your likely diagnosis based on the MRI images provided in Figure 2?

POSSIBLE ANSWERS TO MULTIPLE-CHOICE QUESTION

A. Nasal carcinoma
B. Dermoid sinus
C. Intracranial abscess

CORRECT ANSWER

B.

Extra-axial mass lesion located at the midline of the frontal lobes. The mass appears iso-/irregularly hyperintense in T2W and STIR sequences, heterogeneously T1W and FLAIR iso- to hypointense, and heterogeneously hyperintense in the T2*GE sequence with a thin peripheral rim characterised by 'signal void artefact'. In the T1W post-contrast sequences, the mass is peripherally contrast enhancing. All sequences are described relative to grey matter.

The mass appears to be in contact with the falx cerebri and extends rostrally through a frontal bone defect into the left frontal sinus and then reaching the skin surface.