Title

Increased incidence of thoracic wall deformities in related Bengal kittens

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Summary

Clinical records made during routine vaccinations were compared between populations of Domestic Short Hair (DSH) and Bengal kittens. An increased incidence (12/244) of thoracic wall deformity was detected amongst the Bengal kittens. Deformities detected were: pectus excavatum (5), unilateral thoracic wall concavity (6) and scoliosis (1). Five generation pedigrees were analysed for the affected kittens that showed a high degree of common ancestry indicating the likelihood of a familial cause.

Introduction

A variety of different thoracic wall deformities are described in kittens including pectus excavatum (PE), flat-chestedness, scoliosis, lordosis, kyphosis, pectus carinatum and missing or extra ribs or thoracic vertebrae. However there is a paucity of literature in this area with only 23 cases of pectus excavatum previously reported[1-14] and a single paper on flat-chestedness in Burmese kittens.[15]

In man, PE is the most common thoracic wall abnormality occurring between 1:400 and 1:1000 live births[16,17] with a male bias and is commonly associated with connective tissue disorders such as Marfan and Ehlers Danlos syndromes. The aetiology of pectus excavatum in man is uncertain, but a familial tendency has been found with multiple modes of inheritance suggested.[16] PE can occur as a single entity or associated with other abnormalities and may well be the phenotypic response to a variety of underlying conditions. In many cases thoracic wall deformities are cosmetic but in some individuals the reduction in thoracic volume causes significant respiratory compromise.

The incidence of thoracic wall deformities in kittens is unknown but information from breeders would suggest that they are not an infrequent occurrence. In a study of flat-chested Burmese kittens Sturgess et al.[15] reported an incidence of 3-4% of live Burmese kittens with a suspected genetic component although the mode of inheritance was unclear, the number of kittens with PE in this study was also higher than expected (Sturgess personal communication) and this was considered a

separate clinical entity.[15] Pectus excavatum has been associated with mucopolysaccharidosis VII in cats[18] but the underlying aetiology is unknown. There is a range in severity of deformity with various degrees of respiratory compromise induced by the loss of thoracic volume. The authors have examined multiple Bengal kittens affected by PE or other thoracic wall deformities. Some cases have been asymptomatic while others have had severe respiratory compromise. This report describes the occurrence of multiple, different thoracic wall deformities in a group of related Bengal kittens and tests the hypothesis that the incidence of thoracic wall deformity within this group is greater than that of unrelated Bengals and also that the incidence within the Bengal breed is greater than that of a domestic shorthair (DSH) population.

Material and Methods

Animals

Records of Bengal kittens presented for kitten vaccinations at a single UK practice were examined for evidence of thoracic wall abnormalities as it had been noted that more than one kitten within some litters was affected. A total of 252 Bengal kittens presented for primary vaccination between January 2004 and June 2011. Kitten name, age, sex, weight and results of clinical examination were available for 244 (125 male and 119 female) kittens and these were reviewed. A five generation pedigree was then obtained from the breeders for all affected kittens. The incidence of thoracic wall deformities was then compared to those recorded in 1748 domestic short hair kittens who presented for routine vaccination at the same practice over a similar period and for whom information on the same clinical parameters was available.

All animals (Bengal and DSH) underwent palpation of the thoracic wall as part of their routine clinical examination. Kittens were diagnosed with PE by palpation alone using previously described criteria.[15] In all cases a prominent dorsal deviation of the caudal sternum was palpated resulting in dorsoventral narrowing of the caudal thoracic cavity. Any abnormal findings eg PE were described in

the clinical notes. Cats who presented as dyspnoeic were radiographed and advice only given to asymptomatic cases.

Data analysis

The incidence of thoracic wall deformities in Bengals was compared with non-Bengals as well as the incidence in kittens related to the founder tomcat in the pedigree compared to other Bengal kittens.

Groups were compared using a two-sided Fisher's exact test. Confidence intervals were calculated using Woolf's approximation. Significance for statistical analysis was taken as P value of <0.05.

Results

No thoracic wall abnormalities were recorded in the domestic short hair kittens compared to 12 cases in the Bengal kittens (p <0.0001). Odds ratio (OR) 1053 (95% CI 61-18103). 9 litters had affected individuals with 3 litters having 2 affected kittens present. 5 of the kittens were recorded as having PE, 6 with unilateral thoracic wall concavity (UTC) and one with scoliosis. Kittens in the related pedigree (Fig. 1) were significantly more likely to have thoracic wall deformity than other Bengal kittens (P<0.001; OR 17.8; 95% CI 6.7-2030).

Of the 3 litters with multiple affected kittens, 2 had kittens with UTC only whilst the 3rd litter included one kitten with PE and one with UTC.

PE occurred in 4 female and 1 male kitten (p = 0.37 male vs. female) whereas UTC occurred in 5 male and 1 female kitten (p = 0.07 male vs. female; OR 8.1 [95% CI 0.87 – 74.7]).

Analysis of the 5 generation pedigree (Fig 1) indicated a strong degree of common ancestry. There was no sex bias (6/23 male and 6/30 female) in the affected kittens (p=0.74). There were 41 unaffected kittens within the 9 litters (17 male; 24 female).

All of the Bengal kittens examined were bred by one of two breeders.

All of the affected kittens were asymptomatic at time of initial examination. Three of the five kittens with PE represented with tachypnoea between 4 and 7 months of age. Two of these kittens underwent corrective surgery (external splinting). One kitten survived the postoperative period and is currently asymptomatic (18 months follow up) whereas the second kitten (Fig 2) was severely dyspnoeic on presentation with marked pulmonary hypertension and died of re-expansion pulmonary oedema in the immediate post-operative period, a previously reported complication.[2] The remaining 2 cases of PE were lost to follow up before skeletal maturity was achieved.

The kittens with unilateral thoracic wall concavity (UTC) were variably described in the clinical records as having either “rib cage/thoracic wall defects” or “abnormal proximal termination of ribs”. In all cases the caudal thoracic cavity was laterally compressed and in 5/6 cases this was due to a concavity in the right caudal thoracic wall due to either the absence or abnormal shortening of ribs at this site. UTC did not lead to respiratory compromise in any affected kitten.

Discussion

Pectus Excavatum is an uncommon congenital defect which has been previously reported in cats.[1-14] The unilateral thoracic wall concavity (UTC) described here has not been previously reported. Unilateral lumbarisation of T13 has been previously described[19] but this would not lead to the marked deformities seen in the kittens presented in this report. It has not been possible to radiograph the affected kittens to further elaborate on the nature of these defects and this may be seen as a shortcoming of this paper. The aim of this study was not, however, to describe the deformities seen. The aim of this paper was to test the hypothesis that there is an increased incidence of thoracic wall deformities within the Bengal breed compared to a DSH population and to compare the incidence of thoracic wall deformity in a group of related Bengals to that of unrelated Bengal kittens. The data presented here supports the hypothesis that these deformities are more

common within the examined Bengal population than a DSH population and suggest that there may be a familial tendency.

Assuming that the various thoracic deformities recorded in this group of Bengal kittens represents a spectrum of phenotypic expressions resulting from a similar underlying cause these data strongly suggest a familial association. A previous study has also made an association between an increased incidence of more than one thoracic deformity (PE and flat chestedness) in a specific breed (Burmese) with indications of a familial tendency.[15] These results do not, however, establish that the thoracic wall deformities are necessarily heritable as environmental influences have not been excluded. If the data presented represents a single heritable condition then it is occurring too infrequently to be a dominant mode of expression. Simple recessive inheritance is also unlikely as more affected kittens would also be expected. The data set is too small to make further pedigree analysis but, similar to that suggested by Sturgess *et al*, a complex inheritance is likely with possible incomplete penetrance and environmental influences.

Incidence of PE in man has been estimated at affecting 1:400 to 1:1000 live births.[16,17] Motivations for surgical repair include compromised respiratory function and cosmesis with associated psychological effects.[20] The most commonly performed surgical corrective procedures are modifications of the Nuss procedure which involves placement of a substernal metal bar to correct and splint the deformity.[21]

The most commonly used corrective technique in cats is to use an external body cast with circumsternal/costal sutures to traction the dorsally displaced caudal sternebrae.[1] Risks of this procedure include visceral trauma during suture passage,[1] re-expansion pulmonary oedema[2] as well as the risks inherent to anaesthesia of a juvenile animal with respiratory compromise. Surgical correction is indicated when respiratory function is compromised.[1] Asymptomatic kittens should be monitored closely until adulthood and any evidence of respiratory compromise due to PE should prompt clinicians to consider surgical intervention.

The severity of PE was not detailed in all cases presented here and so we cannot comment on whether or not the degree of PE seen at vaccination (age 9-12 weeks) correlates with likelihood of developing future respiratory compromise other than all kittens with PE for which long term follow up is available went on to develop related symptoms.

This is a retrospective study and as such, Bengal kittens with mild thoracic deformities may have been missed. Thoracic deformities clearly occur in DSH kittens (8/23 cases reported in the literature)[2-14] but the incidence is unknown and depending on its prevalence, could reflect under-diagnosis or under-recording in this study. Both DSH and Bengal kittens were all examined by the same veterinary surgeons and so detection rates of any abnormalities would be expected to be similar between the two groups. In fact, approximately 225 DSH kittens examined in this study would need to have thoracic wall deformities that were not detected or recorded for the incidence of thoracic wall deformities to be similar in the DSH kitten population as in the Bengal kittens. In the data presented there is a tendency to a female bias in the PE cases which is contrary to the experience in humans but this may well be a statistical anomaly given the small numbers in this study. Interestingly, both Bengals with PE previously reported by Yoon *et al* [13] were male and eleven of eighteen (61%) cases of PE reported in the literature to date (for which the gender is known) have been male kittens.

In summary, the data presented here suggest that thoracic wall deformities may be a relatively common potentially familial defect in Bengal kittens. Bengal kittens should have their thoracic wall carefully evaluated during routine clinical examination. Kittens with PE should be closely monitored for signs of respiratory compromise.

Acknowledgements

The primary author would like to acknowledge financial support from the RCVS Trust. The authors would also like to thank the Bengal breeders for providing the 5 generation pedigrees used in this study.

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Conflict of interest statement

Neither author has any personal or financial relationship which could inappropriately influence or bias this study.

Fig1: Five-generation Bengal Pedigree

Fig 2: Radiograph of severely dyspnoeic kitten with marked pulmonary hypertension. This kitten died of re-expansion pulmonary oedema in the immediate postoperative period.