

Estimates of prevalence of hip dysplasia in Golden Retrievers and Rottweilers and the influence of bias on published prevalence figures

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Objective—To estimate prevalence of canine hip dysplasia (CHD) in Golden Retrievers and Rottweilers and identify sources of bias in published reports.

Design—Prospective study.

Animals—200 clinically normal Golden Retrievers and 140 clinically normal Rottweilers between 24 and 60 months of age referred for hip evaluation (group 1) and 93 clinically normal dogs evaluated for Orthopedic Foundation for Animals (OFA) hip certification (group 2).

Procedure—Hip-extended pelvic radiographs from group 1 dogs were screened for CHD. Radiographs were evaluated twice; the first interpretation used an OFA-type subjective 7-point scoring system, and the second included the caudolateral curvilinear osteophyte as an additional sign of degenerative joint disease. The OFA submission rate of group 2 dogs was determined from the number of official reports returned from the OFA.

Results—Prevalence of CHD in Golden Retrievers ranged from 53% to 73% and in Rottweilers ranged from 41% to 69%. Among dogs referred for OFA evaluation, radiographs from 49 (53%) were submitted to OFA. Of submitted radiographs, 45 (92%) were normal; of radiographs not submitted, 22 (50%) were normal. Radiographs with normal-appearing hips were 8.2 times as likely to be submitted to the OFA. Compared with Golden Retrievers, Rottweiler radiographs were significantly more likely to be submitted for OFA certification.

Conclusions and Clinical Relevance—Prevalence of CHD in these 2 breeds may be much higher than previously reported in the United States. Results suggest substantial bias in the OFA database, which causes lower estimates of prevalence of CHD. (*J Am Vet Med Assoc* 2005;226:387–392)

When canine hip dysplasia (CHD) was first described in 1935, it was thought to be a rare condition.¹ By the 1960s, its existence as a pervasive

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orthopedic disease was well established.^{2,3} In 1961, the AVMA Panel on Hip Dysplasia was convened to address disease diagnosis and management.⁴ The Orthopedic Foundation for Animals (OFA) was established as a hip registry in 1966 originally at the University of Pennsylvania to provide a means of applying positive selection pressure in breeding programs and facilitating reduction in disease prevalence. Despite decades of selective breeding with subjective hip scoring, the disease continues to be encountered with high frequency worldwide.⁵⁻⁷

The true frequency of CHD remains unknown. For 2 breeds commonly afflicted with CHD (Golden Retrievers and Rottweilers), prevalence values reported in the literature^{5,8-14,a} range from 22% to 47% in Rottweilers and 23.5% to 55.7% in Golden Retrievers (Table 1). Low values (23.3% and 35.4% for Rottweilers and 23.5% and 30.3% for Golden Retrievers)^{5,8} are derived from OFA data and, to the authors' knowledge, are the only values reported for CHD frequency in the United States for these 2 breeds. Although a disease prevalence of 23% to 35% is quite substantial, it is lower than that reported in all other countries^{10,12-14,a} (with the exception of Norway¹¹) and is empirically lower than would be expected in the United States. An additional OFA report⁹ from 1997 revealed slightly higher frequencies. However, these results were derived from preliminary data collected from screening of radiographs from dogs < 2 years of age. These results contradicted reports from non-OFA sources that indicate high false-negative diagnoses and inability of preliminary hip evaluations to acceptably predict later scores at 2 years of age and development of degenerative joint disease by 1 year of age.^{15,16} It is generally accepted that the proportion of CHD-positive radiographs in the OFA database is artificially low partly because of veterinarians' prescreening of radiographs intended for OFA evaluation.¹⁷ That is, many radiographs of dogs with obvious hip dysplasia are not submitted to the OFA. The OFA does not have a mandatory hip radiograph submission policy; therefore, dog owners have the option of submitting or not submitting their dog's radiographs. In a recent report,⁸ it was stated that prescreening of radiographs submitted to the OFA for preliminary evaluation had little to no effect on the estimated prevalence of HD.

The purpose of the study reported here was to estimate a range of possible values for the true prevalence of CHD in random samples of 2 canine breeds com-

Table 1—Frequencies (%) of canine hip dysplasia (CHD) in Golden Retrievers (GLDR) and Rottweilers (ROTW), as reported in the literature.

Author	Year	Country	GLDR	ROTW
Paster et al*	2004	USA	53–73	41–69
Rettenmaier et al ⁸	2002	USA (OFA)	30.3	35.4
Corley et al ⁹	1997	USA (OFA, preliminary data)	32.6	38.3
Flückiger et al ¹⁰	1995	Switzerland	51	38
Rutkowiak ¹⁴	1994	Poland	ND	41.7
Corley ⁵	1992	USA (OFA)	23.5	23.3
Lingaas and Heim ¹¹	1987	Norway	29	22
Bargai ⁴	1986	Israel	ND	47
Martin et al ¹³	1980	Canada	55.7	ND
Henricson et al ¹²	1972	Sweden	45.4	44.9

*Values obtained from the study reported here, given as a range based on radiographic evaluation with and without the caudal curvilinear osteophyte considered as an early sign of osteoarthritis, which is diagnostic of CHD. ND = Not done. OFA = Orthopedic Foundation for Animals.

monly affected with CHD (Golden Retrievers and Rottweilers) and to explore reasons for discrepancies in reported frequency values. Prevalence estimates from a non-prescreened sample of dogs were intended to reflect a truer estimate of CHD in the United States.

Materials and Methods

Estimation of prevalence of CHD—The study included 340 client- or breeder-owned dogs (200 Golden Retrievers and 140 Rottweilers [group 1]) between 24 and 60 months of age, referred for routine prebreeding hip evaluation between January 1985 and December 1995 and randomly selected from the PennHIP^b database. This sample population was selected to emulate that of other hip registries. Accordingly, dogs with obvious clinical signs of CHD were not part of this cohort because they were not potential breeding animals. Dogs with a prior history of systemic disease or trauma were excluded from the study. All dogs were radiographically evaluated with use of heavy sedation or general anesthesia and the standard ventrodorsal hip-extended radiographic projection.⁴ Hip radiographs were scored by a board-certified veterinary radiologist (DNB) with a standard subjective 7-point scoring system similar to that used by the OFA.¹⁸ For the purposes of this study, radiographs scored as borderline were excluded from the statistical analysis. Each radiograph was scored twice, and evaluations were separated by a minimum of 2 weeks. The first scoring of hip radiographs was based on detection of femoral head subluxation or luxation and osteoarthritis. The second evaluation, performed completely blind to the first, included the aforementioned criteria but also included the **caudolateral curvilinear osteophyte (CCO)** as an additional sign of osteoarthritis. Consistent with OFA scoring, hip joint osteoarthritis was considered pathognomonic for CHD in both evaluations (regardless of whether femoral head subluxation or luxation was present).

Submission rate to the OFA—All dogs of any breed referred to the Matthew J. Ryan Veterinary Hospital of the University of Pennsylvania (MJRVHUP) between January 1991 and December 1992 specifically for OFA certification were identified. Subsequent review of the records of these dogs identified those dogs for which radiographs were actually submitted to the OFA for evaluation. Radiographs were determined to be submitted to the OFA if an official score sheet from the OFA was returned to MJRVHUP. All radiographs, whether submitted or not submitted to the OFA, were evaluated by a board-certified veterinary radiologist at MJRVHUP (DNB) with the standard 7-point OFA-type scoring system. It is important to note that the CCO was not read as an early sign of osteoarthritis in these radiographs. The frequency distribution of hip scores from radiographs sub-

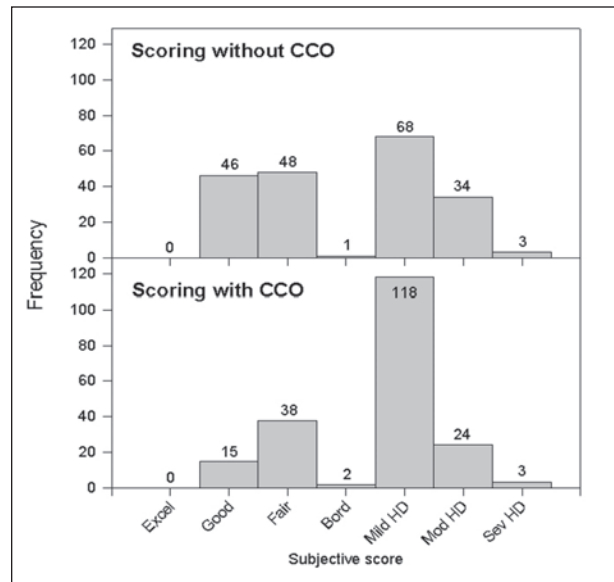


Figure 1—Distribution (frequency [No.]) of subjective, 7-point radiographic hip scores for canine hip dysplasia (CHD) in 200 healthy Golden Retrievers without clinical signs of CHD, as evaluated by a radiologist from the ventrodorsal hip-extended view. Each radiograph was evaluated twice, once with the caudal curvilinear osteophyte (CCO) considered as a sign of CHD and once without use of the CCO. Excel = Excellent. Bord = Borderline. Mild HD = Mild hip dysplasia. Mod HD = Moderate hip dysplasia. Sev HD = Severe hip dysplasia.

mitted to the OFA was compared with the frequency distribution of radiographs that were not. Submission rates by breed were also compared by use of the Fisher exact test. Results are presented as **odds ratios (ORs)** for breed predilection for CHD and relative risk for CHD. To determine the level of agreement between the board-certified radiologist and the OFA, the weighted κ statistic was used. Significance was set at $P < 0.05$, and all analyses were performed with statistical software.^c

Results

Estimation of the prevalence of CHD—One hundred five of the 200 (53%) Golden Retrievers were considered to have CHD when scored according to OFA criteria (ignoring CCO as a sign of osteoarthritis). With the inclusion of the CCO as a sign of osteoarthritis (and therefore diagnostic of CHD), 145 (73%) were scored as having CHD (Figure 1). Fifty-eight of the

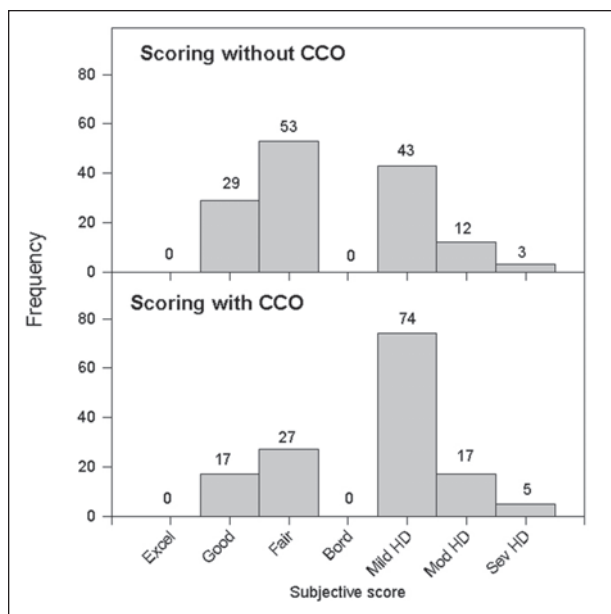


Figure 2—Distribution (frequency [No.]) of subjective, 7-point radiographic hip scores for CHD in 140 healthy Rottweilers without clinical signs of CHD, as evaluated by a radiologist from the ventrodorsal hip-extended view. Each radiograph was evaluated twice, once with the CCO considered as a sign of CHD and once without use of the CCO. See Figure 1 for remainder of key.

Table 2—Frequency of submission of radiographs from 93 dogs consecutively referred for Orthopedic Foundation for Animals (OFA) evaluation and the number of dogs with a passing OFA score. Comparisons were made between Golden Retrievers, Rottweilers, and other breeds combined.

Variable	No. of dogs	No. (%) submitted to OFA	No. (%) with passing OFA score
GLDR	19	4 (21)	4 (100)
ROTW	19	14 (74)	13 (93)
Other	55	31 (56)	28 (90)
Total	93	49 (53)	45 (92)

140 (41%) Rottweilers in the study had CHD when scored without the CCO, and 96 (69%) had CHD when the CCO was considered a sign of CHD (Figure 2). Three Golden Retrievers were scored in the borderline category: 2 dogs when the CCO was included as a sign of CHD and 1 dog when the CCO was not included. No Rottweilers were scored in the borderline category.

Submission rate to the OFA—Between January 1991 and December 1992, 93 dogs representing 32 breeds were referred to MJRVHUP specifically with the owner's intent to send radiographs for OFA certification (19 Golden Retrievers, 19 Rottweilers, and 30 breeds represented by ≤ 5 dogs; Table 2). Of the 93 dogs, hip radiographs for 49 (53%) were submitted to the OFA. Forty-five of the 49 (92%) radiographs submitted received a normal hip score from the OFA. Four of the 19 Golden Retriever hip radiographs (21%) in the sample were submitted to the OFA for evaluation, whereas 14 of the 19 (74%) Rottweiler radiographs were submitted; the difference was significant (OR, 10.5; $P < 0.001$). All 4 Golden Retrievers and 13 of 14 Rottweilers received passing scores. Graphic comparison of the radiologist's (DNB) scoring of submitted

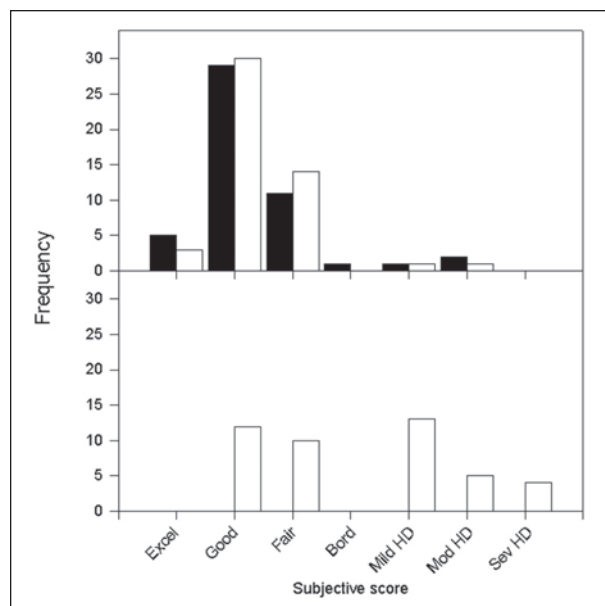


Figure 3—Distribution (frequency [No.]) of subjective, 7-point radiographic hip scores for CHD in 93 healthy dogs referred specifically for Orthopedic Foundation for Animals (OFA) evaluation. Upper histogram—Distribution of scores for 49 radiographs that were eventually submitted to the OFA. Solid bars indicate official OFA scores; open bars indicate scores provided retrospectively by a non-OFA radiologist. Lower histogram—Distribution of scores for 44 radiographs not submitted for official OFA evaluation, as provided retrospectively by a non-OFA radiologist. See Figure 1 for key.

radiographs (excluding the CCO) with official OFA scoring revealed that they shared marked similarity (Figure 3). The weighted κ statistic, however, revealed that agreement between interpretations was low to moderate ($\kappa = 0.56$). The radiographs not submitted to OFA, as interpreted by the radiologist, were 12.2 times as likely to have failing scores, and this difference was significant ($P < 0.001$). Alternatively, radiographs of dogs with normal hips, as judged by the radiologist, were 8.2 times as likely to be submitted for OFA certification ($P < 0.001$).

Discussion

True disease prevalence is difficult to determine. Because it is not possible to survey each member of the entire population, it is necessary to estimate prevalence by studying a representative sample of the population. The sources of error in disease prevalence estimation are many and include case definition (agreement as to what is a positive result indicating CHD and at what age because CHD is a developmental disease), membership bias¹⁹ involving accurate identification of the underlying population of interest (eg, whether we are interested in all dogs, only 1 or a few breeds, dogs that weigh > 30 kg [66 lb], only dogs in the United States, or dogs throughout the world), and selection bias¹⁹ relating to sampling method (whether the sample from the population under observation truly represents the total population). These sources of bias are well defined in statistical references,¹⁹ and each of these factors may contribute to the wide range of CHD prevalence values reported in the literature.

This study attempted to bracket the range of prevalence estimates of CHD in Golden Retrievers and Rottweilers at 53% to 73% and 41% to 69%, respectively. These values are approximately twice the magnitude of the previously reported breed prevalences of CHD in the United States. The causes of such a large disparity are in the described sources of bias (primarily case definition and selection bias) that can affect prevalence estimates. Such sources of bias, to a greater or lesser degree, may have affected our study, previous studies, or both.

It is clear that subjective radiographic evaluation of CHD is complicated. Between- and within-radiologist grading for CHD varies significantly when using standard ventrodorsal hip-extended radiography and a standard 7-point scoring system such as those used in our study.^d That is, a single radiograph evaluated by 2 board-certified radiologists or evaluated by the same radiologist at 2 times will often receive different radiographic scores. For our study, 1 radiologist evaluated the radiographs twice within the range of criteria, which, in the radiologist's estimation, would be considered acceptable radiographic signs of CHD; 1 interpretation was evaluated at the lower or lenient end of the range (ignoring the CCO), and 1 interpretation was at the upper or stricter end of the range (with inclusion of the CCO). The OFA scoring system is subjective, and there are no published detailed definitions of each category. The subjectivity of the 7-point scoring system and the resultant within-radiologist variation explains why some dogs rated as having moderate CHD in the first evaluation were rated as having mild CHD in the second evaluation.

Hip dysplasia grading systems vary widely, can use from 5 to 106 subjective categories for evaluation,²⁰ and may vary in the subjective cutoff point for a passing score. Furthermore, different aspects of radiographic evaluation may be emphasized to varying degrees in different scoring systems. Certainly, the radiographic diagnosis of CHD can be affected by the scoring method, the radiographic criteria used, and the age of the dog at evaluation. Although based on similar positioning and radiography techniques (standard ventrodorsal hip-extended projection), differences in scoring systems used by various hip registry organizations may account for considerable variation in what is considered a positive diagnosis for CHD. A notable example is the criteria for radiographic evidence of osteoarthritis. In previous studies,^{21,22} the presence of the CCO was significantly associated with radiographic signs of osteoarthritis, both contemporaneously and longitudinally. In those studies,^{21,22} dogs with this sign were 7.9 times as likely to have a contemporaneous association with osteoarthritis and were 3.7 times as likely to develop obvious radiographic hip osteoarthritis later in life, compared with those lacking this sign. Some radiologists, including OFA radiologists, do not interpret the CCO as an early sign of osteoarthritis (and therefore CHD) if the other diagnostic criteria for CHD (further evidence of hip osteoarthritis and laxity) are not evident. This is certainly a source of case definition bias. In this report, an upper limit of prevalence for CHD involved inclusion of the CCO as a criterion of hip osteoarthritis.

The sampling method used in our study was to randomly draw a representative group of Golden Retrievers and Rottweilers from a database comprised of potential breeding animals. If the database from which the sample was derived was already inherently biased, the sample would be correspondingly skewed and the prevalence value calculated from it would be inaccurate. The explanation for the lower than expected prevalence values previously reported in OFA data in the United States is thought to derive, at least in part, from selection bias.¹⁷ That is, when owners request radiographic hip evaluation with the intent to submit the radiographs for certification by the OFA, the radiographs may not ultimately be submitted for evaluation. If, in the opinion of the referring veterinarian or others, the hips are of suboptimal conformation or dysplastic, an owner may be less likely to follow through with submission because certification is unlikely to be granted. As a consequence, the pool of radiographs available for frequency analysis in the OFA database is unidirectionally skewed toward normal hip joints, yielding low frequency values derived from the database. It is for this reason that our CHD prevalence estimates for the clinically normal 200 Golden Retrievers and 140 Rottweilers were derived from hip-extended radiographs in a database with mandatory film submission. This selection bias is exacerbated by the case definition bias; specifically, less stringent radiographic criteria will further bias the OFA prevalence values towards lower values.

The degree to which the OFA database is skewed is unknown. In the second part of our study, 93 owners referred dogs specifically for OFA certification and 53% eventually submitted the radiographs. Interestingly, in the small sample of Golden Retrievers and Rottweilers in this group in our study (19 for each breed), there appeared to be breed specificity for radiograph submission rate. Reasons for this breed-specific phenomenon are not clear, although if the frequency of disease is, in fact, much higher in the Golden Retriever breed, prescreening of radiographs would potentially result in a much lower submission rate, compared with Rottweilers. Alternatively, if only 1 in 5 Golden retriever radiographs are submitted, prevalence of CHD could potentially be as high as 80% in that breed. These results, which indicate breed specificity in radiographic submission, do not support the published OFA claim²³ that all breeds were similarly affected by the admitted bias associated with prescreening.

It was not possible to determine exactly what the OFA scores would have been for the radiographs that were not submitted to the OFA. However, when the radiographs not submitted to the OFA were evaluated by a board-certified radiologist, 50% (22/44 radiographs) were deemed to indicate CHD. Specifically, radiographs not submitted to OFA, as read by the radiologist, were 12.2 times as likely to have failing scores when compared with those submitted, and this difference was significant. In contrast, the sample of dogs with normal-appearing hips, as evaluated by the radiologist, was 8.2 times as likely to be submitted for OFA certification. It is not clear why 50% of nonsubmitted radiographs revealed normal hip joints. Breeders obvi-

ously consider factors other than hip conformation in deciding to submit radiographs to the OFA. Although it is not possible to accurately predict OFA score from the radiologist's evaluation, his scoring, without inclusion of the CCO in the subset of submitted radiographs, was graphically similar to OFA ratings. However, the weighted kappa analysis, quantifying agreement of hip scores, only revealed low to moderate agreement because of the low frequency of radiographs submitted from dogs with CHD and the subjectivity of the scoring system itself. The disease prevalences from unsubmitted radiographs were subjectively quite different from the 100% and 93% pass rates for Golden Retrievers and Rottweilers, respectively, derived from radiographs submitted to the OFA. These differences were substantial and not explainable by differences in subjective criteria between the radiologist and the OFA.

These results suggest that the OFA database is biased toward normal hip joints. A report⁸ in 2002 argued to the contrary. However, both an earlier OFA brochure²³ as well as a more recent promotional brochure²⁴ acknowledged that prescreening did "affect the reported frequency of hip dysplasia in each breed and therefore, OFA data under-reports the frequency of dysplasia in the general population for each breed." Whether the bias revealed in our report can explain the approximately 2-fold difference in prevalence values is less clear. The population for which we are attempting to establish prevalence of CHD is similar in character to the population the OFA describes: healthy adult dogs ≥ 2 years of age, with no clinical signs of disease. Most of the dogs in our study came from the eastern part of the United States, which may represent a regional membership bias. Although there may be regional variations in CHD frequencies, such variations are unlikely to account for the 2-fold differential we detected.

Likewise, the fact that the MJRVHUP is a referral center may have influenced which dogs were referred, thus creating a selection bias.¹⁹ Owners or breeders submit dogs for hip evaluation that they consider to be the best of the breed. Without such preselection, it is likely that prevalence of CHD would be even higher than reported in our study or from OFA data. It is also worth noting that a sampling of dogs older than the population studied here would be expected to have a further increase in prevalence of CHD, as confirmed in other studies^{25,e-g} in which hip joint osteoarthritis increases in frequency as dogs age.

Based on results of our study, the prevalence of CHD in Golden Retrievers and Rottweilers in the United States is much higher than was previously reported. The higher estimates of prevalence reported here begin to approach values similar to those predicted by use of the PennHIP method.²⁶ It is imperative to maintain an accurate database of the status of CHD in the United States if we wish to understand the true extent of the disease and track the progress of selective breeding programs purported to provide control of CHD. Even if we were to accept the biased estimates of the OFA, the prevalence of CHD is unacceptably high and we, as veterinarians, should commit ourselves to reducing it by evidence-based means.

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Selected abstract for JAVMA readers from the American Journal of Veterinary Research

Incidence, risk factors, and heritability estimates of hind limb lameness caused by hip dysplasia in a birth cohort of Boxers

Marjan A. E. van Hagen et al

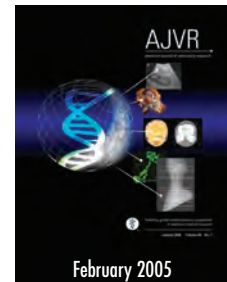
Objective—To determine incidence, risk factors, and heritability estimates of hind limb lameness caused by hip dysplasia in a birth cohort of Boxers.

Animals—1,733 Boxers from 325 litters.

Procedure—Status of Boxers with respect to clinical signs of canine hip dysplasia (cCHD) was registered during an 8-year period. Survival analysis accounted for dogs lost to follow-up. Effective heritability for developing cCHD was estimated by use of a proportional hazard model on the basis of the Weibull distribution. Parametric survival models were developed to identify the influence of potential risk factors.

Results—Cumulative hazard rate for cCHD from 7 weeks to 8 years of age was 8.5%. Dogs that were kept on a floor covered with a slippery material were 1.6 times as likely to develop cCHD, compared with dogs kept on a nonslippery floor. Risk of cCHD doubled in dogs from litters with a high preweaning mortality rate. Dogs that were neutered at 6 months prior to a diagnosis of CHD were 1.5 times as likely to develop cCHD, compared with sexually intact dogs. Dogs > 5 years of age were 1.8 times as likely to develop cCHD, compared with younger dogs. Estimated effective heritability of cCHD was 0.11. In terms of the risk of cCHD in progeny, mean estimated breeding value (EBV) of the 10 best and 10 worst sires was –0.32 and 0.42, respectively.

Conclusions and Clinical Relevance—Registration of Boxers that develop cCHD may provide a strategy for disease prevention. In addition to diagnostic evaluation of radiographs, sire EBVs provide useful information for breeding selection decisions. (*Am J Vet Res* 2005;66:307–312)



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