

Review

A review of literature on the syndrome of arthrogryposis and palatoschisis (S.A.P.) in *Charolais* cattle 1976-1979 (1)

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Summary

Twenty or so papers concerning S.A.P. have been published since 1976 and can be added to the 49 already included in a descriptive bibliography published in 1976.

Important information on the syndrome has been added: the age of the embryo at first appearance has been determined (about 3.5 months). An identifiable anomaly affects the nervous system at the level of the motor end plaque, followed by a disorganisation of muscle tissue. The anomaly presents similarities with a hereditary condition in the mouse known as *muscular dysgenesis* (mdg).

The frequency of the syndrome in purebred *Charolais* in France as well as in Canada has been reported at 0.5 p. 100 among newborn calves. The gene frequency is $q = 0.20$ and penetrance in homozygotes is 0.15 and 0.08 in males and females respectively.

It has been demonstrated that penetrance may appear to be complete as a result of differences in the probabilities of affected calves being reported depending on the numbers occurring in individual herds or sire groups. It is also pointed out that breeding experiments using animals already identified as carriers could result in elevated estimates of penetrance.

Two methods of detecting carrier bulls have been applied: using a test herd of carrier females as in Canada or in the normal course of progeny performance testing pure bred beef cattle as in France. In each case only a small number of bulls can be tested compared with the number required to meet the needs of the breed.

Total eradication of calf losses by S.A.P. is not only theoretically impossible but could even be undesirable, at least from an economic point of view since heterozygous females may be sufficiently more prolific to more than compensate for the low losses resulting from the presence of the gene in the population. Nevertheless, national associations of *Charolais* breeders have other concerns than those associated with the genetics of calf losses, namely commercial considerations.

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To conclude, it would be desirable to continue monitoring the occurrence of S.A.P. calves to avoid confusion with other abnormalities. Continued studies of manifestations of the gene at all levels (histological, histochemical, etc.) and on the etiology of the anomaly are also recommended. Finally, additional investigations of the possible advantage in prolificacy of heterozygous females should be undertaken.

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Introduction

The first scientific publications of the Arthrogyrosis and Cleft Palate Syndrome (S.A.P.) appeared in 1967 (LAUVERGNE et BLIN).

A bibliography prepared by LAUVERGNE and FAUCON (1976) contained 49 papers published during the period 1967-75. Since then some 20 additional articles have been published on this subject which are reviewed in this paper.

Source of the Reports Appearing Since 1975

There is a series of articles describing the investigations conducted at the *University of Saskatchewan*, Saskatoon, Canada supported financially by the *Canadian Charolais Association* (C.C.A.) and the *International Charolais Federation* (F.I.A.E.R.B.C.): QUANTZ (1976), RUSSEL (1976a), RUSSEL (1976b), ANONYMOUS (1977), NIELSEN *et al.* (1977), RUSSEL (1977), ANONYMOUS (1978), RUSSEL (1979).

There have been reports from Edmonton, Alberta (GOONEWARDENE and BORG, 1976) and from Saskatoon (HOWELL and LAUVERGNE, 1978) the latter dealing with a survey of *Charolais* breeders in Saskatchewan.

In France, LEFORT *et al.* (1977) have analyzed the results from a series of 10 tests of *Charolais* bull-calves, while RIEGER (1979) has presented the first results of a French neurobiological study of the neuromuscular system of S.A.P. calves.

Reports from Great Britain (LOGUE *et al.*, 1977, JACKSON, 1978), Belgium (HANSET *et al.*, 1978) and Australia (HARTLEY and WANNER, 1974, overlooked in the listing by LAUVERGNE and FAUCON, 1976) have recorded cases of S.A.P. appearing in *Charolais* in these countries.

Included also is a report on the Canadian situation requested by F.I.A.E.R.B.C. (LAUVERGNE, 1976), on the prospects of eradication (LAUVERGNE and HOWELL, 1977) and on the situation in the United States of America (LAUVERGNE, 1979).

Description of the syndrome (S.A.P.)

These last years have seen marked progress in an understanding of the histopathology of S.A.P. beginning in 1976 with work by RUSSEL on which he reported at the *Monterrey Convention* (RUSSEL, 1977) and described in detail in his Ph. D. thesis of 1979.

Using a part of the C.C.A.'s experimental herd assembled at Goodale near Saskatoon, RUSSEL obtained embryos displaying various stages in the development of the syndrome. In addition he studied S.A.P. calves at birth.

From this study it was possible to determine the prenatal age of the embryo at which the syndrome could first be detected (3.5 months) and the kind of abnormalities present at this age: retraction of axons from the post-synaptic membranes.

It appears that a change in the differentiation of neurons could be the cause of muscle hypotonia and the accompanying leg deformities observed in S.A.P. calves.

In the muscle of newborn S.A.P. calves, structural abnormalities are evident at the level of the neural plaques accompanied by faulty differentiation of muscle fibers.

The characteristics of nerve-striated muscle interaction in the S.A.P. calf have been specified by RIEGER (1979) and RIEGER *et al.* (1980). A detailed parallel study in a neuromuscular mutant in the Mouse (« *Muscular dysgenesis* » *mdg*) has shown similarities in the morphological expression of the neuropathologic impairment: the atrophic muscular fibers present a disorganized myofibrillar structure (Z stria), the innervation networks are deeply desorganized as shown by tests of metallic impregnation of nerves, with a lot of contacts with an immature aspect and an abnormal distribution of the innervation (RIEGER and PINÇON-RAYMOND, 1980).

There are also defects of biochemical nature: in the Mouse the biosynthesis of molecular forms of acetylcholinesterase is quite incomplete (as NAWROT, 1976; NAWROT *et al.*, 1979 have also pointed out). In the S.A.P. calf the mechanisms of localisation and restriction of the specific molecular forms of innervation are abnormal. The functional receptor of acetylcholine (neurotransmitter of the neuromuscular synapse) shows an aberrant distribution in the *mdg* mouse as well as in the S.A.P. calf.

The irregular character of the affected muscles has been stressed by RIEGER (1979). The choice of abnormal cases examined by RUSSELL (1979) (all cases has a cleft palate) must not detract from the fact that there are frequent cases where the animal has analogous muscular and tendinous abnormalities but lacks the cleft palate and can be classed as true S.A.P. (LEIPOLD *et al.*, 1969, GIROUD, 1975, LAUVERGNE, 1975).

Behaviour and frequency of the S.A.P. gene

Studies already cited by LAUVERGNE and FAUCON (1976) such as that of LAUVERGNE (1975), established that the S.A.P. condition was hereditary, monofactorial, and caused by an autosomal recessive gene. Although in France the gene seems to have incomplete penetrance, it may be complete in certain cases, at least in Canada.

In France this result has been confirmed beyond doubt from an analysis of beef cattle test data obtained from purebred animals (LEFORT *et al.*, 1977); estimations of penetrance were 0.15 for males and 0.09 for females and the gene frequency was calculated to be 0.20.

Similar frequencies of the abnormality were revealed by a survey among *Charolais* breeders in Saskatchewan (HOWELL and LAUVERGNE, 1978) after allowance was made for percentage of *Charolais* breeding in the crossbreds. This would indicate that the penetrance of the gene is not influenced significantly by crossing with other breeds.

The first explanation of the almost total penetrance estimated from data obtained from cattle breeders such as reported by NAWROT (1973) in Canada or HANSET *et al.* (1978) in Belgium derives from the fact that inclusion of the proband (the first observation) strongly biased the date because of its variable probability of observation: escaping inclusion when only one abnormal appears but certain to be included when several abnormal calves appear in a short time span within a herd.

This explanation could nevertheless hardly be applied to account for the mendelian segregation compatible with complete penetrance of the recessive homozygote observed in the experimental crosses conducted at *Kinsella* (BERG and GOONEWARDENE, 1974) or at *Goodale* (NIELSEN *et al.*, 1976).

Two comments can be made:

— it is possible that some normal appearing breeding animals may be homozygous recessive as demonstrated by BERG and GOONEWARDENE (1976),

— the use of female carriers of the S.A.P. gene, detected as such because they have already produced an abnormal calf may well lead to an increased penetrance. That being the case, it may be wise to exert some selection against the tendency of certain carrier females to increase the penetrance of the S.A.P. gene when their offspring are homozygous recessive.

The problem of eradication

At Harrogate, a C.C.A. report (ANONYMOUS, 1978) emphasized that the histopathological studies in Canada do not allow distinction to be made between the heterozygous carrier and the homozygous normal animals by a simple examination of the living animals.

But it does appear that the works of RUSSEL (1979) and RIEGER *et al.* (1979) could offer orientations for the search of some biochemical or morphological means of detection.

The only method for the detection of normal appearing carriers (hetero or homozygous) remains a breeding test. Detection and removal of the carriers from the breeding population has so far been undertaken only in two countries and by different methods.

— In Canada, the C.C.A. set up an experimental herd of normal appearing carriers. As already stated, a part of this herd was used by RUSSEL (1976) to produce the abnormal embryos used in his studies. Sires to be tested were mated to these presumed heterozygous cows.

Apparently penetrance is very nearly complete (refer to the foregoing remarks concerning augmentation of penetrance in descendants of cows having already produced a defective calf), so it would only be necessary to perform a few matings to identify the genotype of a bull with reasonable probability of not having made an error (QUANTZ, 1976).

— In France, for pure bred bull testing, the frequency of the gene in the entire female population is used ($q = 0.20$) which is high enough but it doesn't have as high a penetrance as in the experimental herd in Canada. Also, the number of calves required for equal precision is much higher (about ten times). According to LEFORT *et al.* (1977), the probability of nondetection of a heterozygous carrier with 150 progeny is 0.16.

The advantage of the French method over the Canadian is the very low cost since the detection of abnormals is made in the ordinary routine of progeny performance testing. However, these two methods permit the testing and detection of only a small number of males each year, and in populations where artificial insemination is not used to any extent, at least in seed stock herds, they are not effective for eradication of the gene.

In fact, as concluded by LAUVERGNE and HOWELL (1977) in their report to F.I.A.E.R.B.C. it can be asked whether eradication is possible or even desirable.

To the first question, the reply is in the negative. Neither of the two methods examined above is capable of detecting all the carrier males in a breed. What's more, if the French method was rigidly applied it would become less and less effective because, as the gene frequency decreased in the female population, there would be required larger and larger numbers of progeny from a bull to eventually declare him a non carrier with the same high level of probability. In the event of there being a selective advantage for the heterozygote, eradication would be theoretically impossible (LEFORT and LAUVERGNE, 1974).

The second question can also be answered in the negative. For one thing the economic loss by S.A.P. is very low (0.5 p. 100 of newborn calves compared to total losses from all causes currently about 10 p. 100), and is probably less than the cost of an intensive eradication program. Secondly if the observations of BERG and GOONEWARDENE (1974) are accepted (that heterozygous females were more prolific than homozygous normal females), the losses caused by the abnormals would be less than the gains resulting from the heterosis phenomenon (3.3 p. 100 additional newborn calves at a gene frequency of $q = 0.20$).

However, there are other reasons for undertaking an eradication program. Commercial considerations may dictate it, especially in North America where the competition among beef breeds is very keen. This partially explains the program undertaken by the C.C.A. to prove that nothing is neglected in breed improvement. Reasoning from the same position of commercial competition, authorities in the *American Charolais Association* (A.I.C.A.) have on the other hand chosen to systematically ignore the very existence of the anomaly, an attitude in line with the conclusions of a genetic study by LAUVERGNE (1979).

Conclusions

Even if it is recommended to the various *Charolais* associations that the anomaly (S.A.P.) be ignored in their testing and selection programs, it would be desirable for them to routinely monitor cases of S.A.P. if only to be assured that it is truly S.A.P. and not an anomaly caused by a different gene or even a virus. In any case, the study of S.A.P. in all its manifestations at the histological and histochemical levels, is far from complete and must be pursued logically.

The fundamental neurobiologic studies of neuromuscular system in the S.A.P. calf could be developed with profit using the experience from the study of mutants in the Mouse (the already quoted *mdg*; RIEGER, 1979), but also of variants in Man such as « *Arthrogryposis multiplex congenita* » studied by BANKER *et al.*, 1957. This type of studies could not only bring new views on the neuro-muscle contact and its stabilization but also new contributions to the knowledge of biochemical parameters in these pathological cases. It could even be possible in the future to detect normal overlap carriers by checking light abnormalities of some of these parameters in the living population of *Charolais* calves (RIEGER *et al.*, 1980).

It would be equally desirable to continue on-farm studies of the frequency of the syndrome in the pure breed and its crosses to confirm the values already reported in France and Canada.

Chronological bibliography of S.A.P. papers 1976-1979

The numeration of the papers comes in the succession of the numeration of the 1967-1975 article given by LAUVERGNE and FAUCON (1976). The omitted article by HARTLEY *et al.*, 1974, has been put at the beginning, with a bis number.

1974

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1976

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1977

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1978

- (63) ANONYMOUS, 1978. *Arthrogryposis.* In «Delegates meeting» XVth World *Charolais* Convention, Harrogate, Yorksh., 17-9-78, 4.
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In Memoriam

Last November 1979, G. LEFORT Professor of Mathematics at the *Institut National Agronomique Paris-Grignon* (France) has suddenly disappeared.

His contribution to the study of S.A.P. in *Charolais* is noticeable. First he helped us in

establishing the equation for measuring gene frequency and penetrances in the French *Charolais* population. He then contributed to estimate these parameters in the French population as well as in the Saskatchewan one.

He gave also the basic principle for the law of eradication of gene in the case we are interested in.

Without him the difficult statistical situations we faced in the study of S.A.P. as well in France as in Canada could certainly not have been so quickly solved.

Résumé

Synthèse des travaux relatifs au syndrome d'arthrogrypose et de palatoschisis (S.A.P.) chez les bovins Charolais 1976-1979

Depuis 1976, une vingtaine d'articles se sont ajoutés aux 49 déjà cotés dans une bibliographie signalétique publiée en 1976.

Des progrès importants dans la description du syndrome sont notés : l'âge d'apparition chez l'embryon est déterminé (autour de 3 mois et demi); l'anomalie de départ frappe le système nerveux au niveau de la plaque motrice. Il s'ensuit une désorganisation du muscle. L'anomalie est fort semblable à une affection héréditaire de la Souris, la *muscular dysgenesis (mdg)*.

La fréquence de l'anomalie en race *Charolaise* pure, aussi bien en France qu'au Canada, s'établit à 0,5 p. 100 des veaux naissant. La fréquence du gène serait $q = 0,20$ et les pénétrances chez les homozygotes respectivement 0,15 chez les mâles et 0,09 chez les femelles.

On a expliqué que, dans certains cas, la pénétrance peut paraître totale à cause de différences dans les probabilités d'enregistrement des fratries selon le nombre d'anormaux apparus, et que, dans le cas de croisements à partir d'animaux porteurs détectés d'après leur descendance, une certaine sélection a pu jouer pour augmenter cette pénétrance.

Deux méthodes de détection des mâles porteurs du gène ont été expérimentées : au Canada, à l'aide d'un troupeau expérimental de femelles porteuses, en France, dans le cadre du testage viande en race pure. Dans les deux cas, un nombre infime de taureaux a pu être testé par rapport aux besoins de la race. L'éradication de la tare, matériellement impossible, semble également théoriquement inutile, du moins économiquement parlant car les femelles hétérozygotes pourraient être plus prolifiques, ce qui compenserait, et au-delà, les faibles pertes dues à la présence du gène dans la population. Cependant, les associations peuvent tenir compte d'autres considérations que des considérations purement économiques et génétiques pour orienter leur action, en particulier des considérations commerciales.

Pour conclure, il est souhaitable que l'on continue de repérer les veaux S.A.P. d'une manière routinière pour éviter de confondre avec d'autres possibles anomalies. Les études sur les manifestations à tous les niveaux et sur l'étiologie pure de l'anomalie sont également préconisées. On aimerait également avoir des précisions supplémentaires sur l'avantage de prolificité des femelles hétérozygotes.

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