TITLE: The efficacy and safety of prophylactic cryotherapy in preventing retinal detachment in type 1 Stickler syndrome

AUTHORS (LAST NAME, FIRST NAME): Fincham, Gregory S.; Pasea, Laura; Carroll, Christopher; McNinch, Annie M.; Poulsen, Arabella V.; Richards, Allan J.; Scott, John D.; Snead, Martin P.

2. Centre for Applied Medical Statistics, University of Cambridge, Cambridge, United Kingdom.
3. School of Health and Related Research, University of Sheffield, Sheffield, United Kingdom.
4. Department of Pathology, University of Cambridge, Cambridge, United Kingdom.
5. Regional Molecular Genetics Laboratory, Cambridge University NHS Foundation Trust, Cambridge, United Kingdom.

Commercial Relationship(s) Disclosure:
Gregory Fincham: Commercial Relationship: Code N (No Commercial Relationship)
Laura Pasea: Commercial Relationship: Code N (No Commercial Relationship)
Christopher Carroll: Commercial Relationship: Code N (No Commercial Relationship)
Annie McNinch: Commercial Relationship: Code N (No Commercial Relationship)
Arabella Poulsen: Commercial Relationship: Code N (No Commercial Relationship)
Allan Richards: Commercial Relationship: Code N (No Commercial Relationship)
John Scott: Commercial Relationship: Code N (No Commercial Relationship)
Martin Snead: Commercial Relationship: Code N (No Commercial Relationship)

ABSTRACT BODY:
The efficacy and safety of prophylactic cryotherapy in preventing retinal detachment in type 1 Stickler syndrome

Purpose: To evaluate the efficacy and safety of 360° prophylactic cryotherapy (360PC) according to a standardized protocol and rationale in preventing retinal detachment (RD) in type 1 Stickler syndrome patients.

Methods: A retrospective cohort study. Four hundred and eighty seven type 1 Stickler syndrome patients with both eyes available for study were allocated to a prophylaxis group (bilateral 360PC) and compared to a control group (no 360PC), or a mixed group (unilateral RD with subsequent bilateral 360PC) and compared to a mixed-control group (unilateral or bilateral RDs with no 360PC) by adjusted multivariate Cox regression and age-matching analyses. All eyes from all patients were also analysed individually to compare the risk of RD with and without prophylaxis, in addition to randomly selecting one eye from each patient for repeated comparison. The main outcome measures were time to RD (including failure of 360PC requiring retinopexy with or without surgery) and side effects resulting from prophylactic treatment.

Results: The control group (n=194) had a greater than seven-fold increased risk of RD compared to the prophylaxis group (n=229) (hazard ratio [HR] = 7.4; 95% confidence interval [CI]: 4.5 – 12.1), with the age-matched control group (n=165) having a five-fold increased risk compared to the age-matched prophylaxis group (n=165) (HR = 5.0; 95% CI: 2.8 – 8.8). Similarly, the mixed-control group (n=104) had a greater than ten-fold increased risk of RD compared to the mixed group (n=64) (HR = 10.3; 95% CI: 5.0 – 21.4), with the age-matched mixed-control group (n=39) having a greater than eight-fold increased risk compared to the age-matched mixed group (n=39) (HR = 8.4; 95% CI: 3.3 – 21.6). Analysis of all individually analysed eyes that received no prophylaxis (n=452) indicated a greater than ten-fold increased risk of RD compared to those that received 360PC (n=522) (HR = 11.7; 95% CI: 7.9 – 17.2), with a greater than fifteen-fold increased risk calculated when one eye from each patient was randomly selected (HR = 15.4; 95% CI 8.1 – 29.3). Reported side effects from prophylactic treatment were minor and short-lived.
Conclusions: All analyses indicate that prophylactic retinopexy delivered according to this standardized protocol is safe and markedly reduces the risk of retinal detachment in type 1 Stickler syndrome.