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The role of athletics in the life of the hemophiliac

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by

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To the Children who Suffer from Diseases of the Blood.

Their Courage in the Face of Serious Illness Continues to Inspire Those of Us Who Have the Privilege of Working with Them.

and

To My Parents

ACKNOWLEDGEMENTS

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I. INTRODUCTION

The hemophilias are a group of inherited coagulation defects caused by the functional deficiency of various plasma protein clotting factors. These disorders are transmitted through a sex-linked recessive Mendelian pattern, with usually unaffected female carriers passing the disease almost exclusively to their sons. The rate of mutation for this gene is quite high, with 40% of all hemophiliacs having no family history whatsoever. The disease is rare, affecting only two out of every 100,000 males.

There are two main types of hemophilia, namely classical hemophilia, or hemophilia A, and Christmas disease, or hemophilia B. In hemophilia A the missing plasma protein is the coagulant portion of Factor VIII, or antihemophiliac factor, while in hemophilia B it is Factor IX, or plasma antecedent component. Hemophilia A is by far the more common, representing about 80% of all hemophilic patients. Most of the remaining patients suffer from Christmas disease, with an occasional case of an autosomally recessive Factor XI deficiency. The clinical manifestations of the diseases are identical.

Based upon the actual degree of factor deficiency, the hemophilias may be classified as either mild, moderate or severe. Severe hemophiliacs have less than 1% of normal factor activity while moderate and mild hemophiliacs have 1-5% and 5-25%, respectively. In spite of this, the clinical course of the disease is erratic and unpredictable,

with only general correlation to factor levels.

The disease has been known since the civilizations of ancient Egypt, and has long been the subject of misconception, mystery and folklore. Although often referred to as the "disease of the Hapsburgs," no scion of that family ever bore its mark. Queen Victoria passed the gene throughout the royal families of Prussia, Spain and Russia. Her great-grandson, the Tsarevich Alexis of Russia, was probably the most famous hemophiliac, but the relationship between him, his mother and the disease was, in essence, little different from that played out in thousands of less powerful and glamorous families. In her grief and despair, his mother reached out to the Siberian peasant and supposed miracle-worker, Gregory Rasputin.[1] As Lubert Stryer has said, "no man knew less about molecular disease, no man ever profitted more from it."[2] The rest, as is said, is history.

Clinically, the disease does not represent the popularly believed ever-present danger of exsanguination. Hemostasis following an external laceration is largely a vascular-platelet phenomenon, and simple pressure will usually stop the bleeding. It is the internal bleeding, rather, which poses the real problem. The two most feared complications of hemophilia, bleeding into the cranium and bleeding into the soft tissues surrounding the air-way, represent truly life-threatening situations. A patient could also hemorrhage into the abdomen or retroperitoneal space with disastrous consequences. Fortunately, these potentially fatal

occurences are not common. The major clinical manifestations are soft tissue hematomas and hemarthroses, or bleeding into the joints. Although in mild and moderate hemophiliacs these bleeding episodes are usually related to trauma, in a severe hemophiliac they frequently occur spontaneously. The joint most commonly affected is the knee, followed in order by the elbow and ankle. The joints of the hand or vertebrae are almost never affected. The blood has a destructive effect on the joints, and repeated episodes can lead to scarring, loss of motion, muscle wasting, arthritis and eventual crippling. While this progressive arthropathy was inevitable a generation ago, advances in factor replacement and physical therapy have led to a much more favorable outlook for today's hemophiliacs.

In the past, strict immobilization was used following a hemarthrosis, but current therapeutic doctrines call for an early return to mobility, combined with specific joint-preserving exercises. Because of this aggressive use of physical therapy, many patients are able to escape the severe crippling residua of these bleeding episodes.

These advances would not have been possible without the discovery, in 1965, of cryoprecipitate. Developed by the late Dr. Judith Pool while working at Stanford, this represented the first concentrated form of factor VIII.[3] Prior to this time, the only sources of the missing clotting factors had been whole blood or fresh-frozen plasma(FFP). The levels of factor VIII were so low in these unconcentrated preparations, however, that in order to obtain reasonable circulating

levels of the missing factors, one was forced to risk circulatory overload by transfusing enormous volumes of the FFP. Now with the discovery of cryoprecipitate, higher plasma levels could be achieved with smaller volume administrations. It was not long before even more highly concentrated preparations were available, preparations that were also easier to store and reconstitute.

In the late 1960's a new concept in the treatment of hemophilia gave the patient even more freedom: home-therapy. The patient or his parents now assess the transfusion needs at the first sign of bleeding and, if necessary, reconstitute and administer it themselves. Patients as young as seven years of age have been taught to successfully treat themselves. In the event of a severe bleed, the patients are taught to contact a physician. This approach has freed the hemophiliac and his family from their geographical restraints and has made possible experiences such as travel and summer camp. In many ways, the hemophiliac is able to lead a normal life.

Unfortunately, there are still major problems. Intracranial hemorrhage continues to be a major cause of death in hemophiliacs. Secondly, approximately 10-15% of hemophilic patients develop an inhibitor to factor VIII. Inhibitors are antibodies of the Ig G class which react with and destroy the transfused clotting factor. As a result, these patients do not have many of the treatment options available to other hemophiliacs.

Two other problems facing many hemophiliacs are directly related to transfusion therapy: liver disease and cost. With

the commercial production of factor concentrates from pooled plasma, contamination with hepatitis antigen is unavoidable. Concerning cost, the average expense of treating a single hemorrhagic episode is over $200. For patients without adequate third-party coverage, this represents a staggering financial committment. Over twenty states have passed legislation providing specific assistance to these patients, but more needs to be done.

Finally, despite the recent advances in therapy, many hemophiliacs continue to be over-protected and sheltered from the activities and experiences of a normal childhood--challenges which they are medically prepared to meet. One important aspect of such a normal childhood is recreational activity. This paper will review the concepts, attitudes and recommendations in the literature concerning participation by hemophiliacs in sports and games. The results of a study of actual sports participation by boys with hemophilia will then be reported.

II. LITERATURE REVIEW

In reviewing the literature, it becomes apparent that little has been written directly addressing the issue of athletic participation for hemophiliacs. Several authors touch on this issue while pursuing other themes, giving valuable insights from different perspectives. The pertinent literature of the psychiatric and physiotherapeutic disciplines will be examined first, followed by a review of the few articles which focus more directly on the question of the proper role of athletics in these patients.

In 1961 Austin et al wrote on various aspects of physical therapy which could be useful in the treatment of residual joint damage secondary to chronic hemarthroses.[4] At that time, swimming was encouraged as the only safe sport for hemophilic patients. By 1974, with the availability of factor concentrates, the thinking of those involved in this field had changed dramatically. Writing in that year Boone stated that "patients of all ages are urged to be as physically active as possible."[5] In particular she recommended swimming, bicycling, tennis, hiking and gardening.

In 1975, while stressing the importance of the prevention of permanent musculo-skeletal damage, Shelby Dietrich encouraged participation in age-appropriate recreational activities.[6] It was her opinion that a modified program of physical education was important for school-aged youngsters with contact sports such as football, basketball and wrestling being avoided. Swimming, tennis, hiking and

gardening were again encouraged. These authors viewed such activities as safe extensions of the patients' formal exercise programs, with their chief purpose being the rehabilitation of chronically damaged joints.

Another beneficial aspect of physical therapy and recreational activity was mentioned by Weissman in an extensive review in 1977.[7] Although unable to document this conclusively, he stated that "there is a general feeling among people who work with hemophiliacs that those patients who maintain good general levels of physical fitness and are well muscled have fewer spontaneous hemorrhages." Deductively, it would seem that a joint supported by adequate musculature would be better able to withstand the everyday trauma of walking, climbing stairs and minor recreational activity. To that end, he recommended swimming, jogging, bicycling, tennis, golf, dancing, croquet, ping-pong, fishing and shuffleboard. Football, wrestling, basketball, volleyball, roller skating, horseback riding, jumping rope and trampoline were discouraged. He did acknowledge, however, that there was some disagreement between physicians over lists of acceptable recreational activities.

As late as the early 1950's hemophilia was considered by at least one author to be the disease least likely to involve a psychosomatic component.[8] By the early 1960's, however, several investigators had begun to examine the role of emotional factors in the genesis of bleeding episodes. This work stimulated broader investigations into

the psychiatric aspects of hemophilia in general.

The first to suggest a possible relationship between emotional factors and bleeding episodes was Paul Poinsard in his presentation of a short paper before the National Hemophilia Foundation in 1956.[9] He reported that it was the general impression of a group of his patients and their parents that emotional stress was more conducive to bleeding than emotional tranquility. Although he made no formal attempt to test this hypothesis, his observations were an important contribution to a fledgling discipline.

Browne, writing in 1960, reported the results of his studies on 28 hemophiliacs and their families.[10] Through his interviews, Browne discovered that trauma was not as important a factor in precipitating bleeding as he had presupposed. In fact, there were several reported instances of severe trauma without subsequent hemorrhagic episodes. Instead, the patients reported variations in their tendency to bleed, an increased tendency being associated with times of anxiety, anticipation or other emotional stress.

Concerning the personality patterns of the patients, Browne found most of the children to be outwardly passive and docile, though with occasional subtly rebellious episodes. The mothers' reactions to their sons' disease were invariably characterized by extreme guilt, and they saw themselves as the only effective protector of the child. The fathers' reactions varied; some were remote and aloof, others anxious, denying or relieved that they were not genetically involved.

Browne's findings prompted further investigation in

this area. Two researchers from Cleveland, David Agle and Ake Mattson, explored a wide range of psychiatric issues in these patients, publishing an important series of papers in the mid and late 1960's.

In 1964 Agle reported the results of his inquiry into two broad areas: 1) the effect of psychological factors on the clinical course of the disease and 2) the effect of the illness on the developing personality.[11] Regarding the former issue, Agle confirmed the work of Poinsard[9] and Browne[10], finding several instances of so-called spontaneous bleeding associated with emotional stress. Even more interesting, however, was his report of marked clinical improvement in eight patients following their conversion from a passive, dependent life-style to a more aggressive independence.

Addressing the second question, Agle noted the importance of familial dynamics as they relate to the effect the disease may have on the hemophiliac's personality. In particular, he noted that the mothers, acting out of their own guilt and anxiety, tended to seriously over-protect the children, unreasonably limiting their activity. The intense nature of the mother-son relationship then often forced the father into the role of an apparently uninterested bystander. This extreme over-protection, in the author's opinion, often led to one of two extremes of behavior. Either the child accepted the views of his mother and led an anxious, passive life, or he rebelled, engaging in risk-taking activity secondary to either denial or a counter-phobic response. To help prevent this, Agle

recommended that, among others, efforts be made "to encourage healthful activity and reasonable aggressive pursuits" for the hemophilic child.

A year later Mattson reported on the psychosocial adjustment of the young hemophiliac.[12] He found that most of the children and their parents showed a positive adjustment to the disease, with the parents allowing their sons participation in swimming, baseball, fishing and a variety of other athletic events. He stated, "these ten well-adjusted patients have tended to participate in a variety of sports as they became less hampered by their illness." Mattson also confirmed Agle's[11] observation of an improved clinical course following the change from an inactive, fearful overprotected life-style to a more active independence. This marked increase in physical activity was accompanied by a significant decrease in hemorrhagic episodes.

Mattson felt that the parents, particularly the mother, played an essential role in the development of a healthy personality in their sons. Moreover, he stressed the need to rear the hemophilic child as normally as possible. The few patients in his study who did display a poor adjustment were the subjects of maternal overprotection, displaying either passive-dependent behavior or rebelliousness.

Publishing together in 1968, Agle and Mattson discussed the adaptive mechanisms which well-adjusted hemophiliacs use to come to terms with their disease.[13] Chief among these were intellectual activity, emotional expression and

motor activity. In exploring the last of these, they stated that "allowing reasonable motor activity to discharge normal aggression often increases the boy's sense of masculine well-being." They warned that it is often important to emphasize to the parents that in spite of the most stringent precautions, some hemorrhagic episodes are certain to occur.

In discussing this issue they also clearly stated the importance of the role of the father in the total care of the child. The fathers of the well-adjusted patients joined their sons in several recreational activities including swimming, golf, baseball, tennis, weight-lifting and touch football. These activities led to surprisingly few bleeding episodes, a result they postulated of the protective effect of well-developed musculature on the joints. Although this protective effect has never been proven in a controlled study, the same impression was expressed by another author earlier in this review.[7] Finally, Agle and Mattson recommended that as the child matures, he be allowed increasing responsibility in his choice of appropriate physical activity.

In the same year Garlinghouse and Sharp, in a statistically rigorous study, looked at the relationship between a hemophiliac's self-concept, family stress and bleeding episodes.[14] As has been discussed, it had long been believed that hemophiliacs suffered more spontaneous bleeds during times of emotional stress. These investigators sought to statistically document this, while also exploring the

relationship between self-concept and hemorrhagic episodes. They found that there was indeed an increase in bleeding episodes in times of stress, although it was not statistically significant. They also discovered an inverse relationship between self-concept and the tendency to bleed. In other words, the stronger the hemophiliac's self-concept, the less spontaneous bleeding observed.

A dissenting opinion on the generally accepted pattern of maternal over-protection was registered in 1976 by Steinhausen.[15] He found that hemophilic boys did not often perceive their mothers as being overly protective. Although this may not reflect accurately on actual maternal behavior, he felt that the stereotype of maternal over-protection was the product of deductive psychodynamic thinking rather than precise clinical observation.

Jonas and Wincott, in separate extensive review articles published in 1977, essentially reiterated the basic tenets put forth by earlier researchers.[16,17]

It was first reported by Rizza in 1961 that the circulating level of Factor VIII in the blood of normal subjects was increased by strenuous muscle exertion.[18] The mechanism of this increase was not understood, but he later showed that it did not represent the release of Factor VIII from the spleen, as had been suggested.[19] A recent study by Turner,et al, again confirmed the rise in coagulant activity of Factor VIII in normal individuals following exercise.[20] This study also examined two individuals with mild hemophilia, reporting an increased Factor VIII level in only one of the two patients

following exercise. The patient who did not show an increased Factor VIII level was felt not to have exercised as strenuously, as determined by 0_2 consumption. Although this information is sketchy at best, the work done in this area has been used as a possible explanation for the decreased frequency of bleeding episodes seen in many hemophiliacs who lead active lives.

In the years since 1970, a small body of literative has come into existance which deals more directly with the issue of hemophiliacs and recreational activity. Butler and associates were the first to report a summer camp experience for hemophiliacs, stating only that the week was a social, medical and recreational success.[21] The camp mixed hemophilic and non-hemophilic children, and was staffed by a hematologist and recreational therapist on a 24-hour basis. The 15 hemophiliacs, staying for a total of 20 camper-weeks, required a total of 17 transfusions during the experience, but there were no serious incidents reported.

In 1975 Seeler reported her experience with an Illinois camp designed specifically for hemophiliacs.[22] Again employing continuous medical coverage, this camp also used para-medical personnel in other staff roles. Boys with inhibitors were not accepted at this camp. Activities included swimming, canoeing, sailing, fishing, archery, horseshoes, volley ball, tennis, badminton, trampoline, nature walks, shuffle board, ping pong and whiffle ball. Combined with these recreational activities, the campers who expressed an interest were given instruction

in self-infusion. She concluded her report wirh the assessment that "by participating in sports where they can safely compete, the boys increased their self-esteem through actual accomplishments. Our experience suggests that physicians and their parents have been overly protective . . ."

Two years later Seeler again reported on the camp, this time stressing behavioral benefits gained from the camping experience.[23] In describing the problem she stated, "Even though effective infusion therapy is now readily and rapidly available, we were surprised at the tremendous amount of parental over-protection that still exists." In fact, she felt that the problem was probably much worse than actually observed, in that the parents of over 50% of all age-appropriate hemophiliacs in Illinois had not allowed their sons to attend the camp. It should be noted that no camper was refused for financial reasons.

During the camping experience Seeler noted a marked decrease in self-pity. Several of the boys reacted to the new situation by being either overly timid or manipulative, necessitating a firm disciplinary approach to settle interpersonal disputes between campers. Although risk-taking behavior has been widely described in the literature, Seeler reported very little of this during the camp.

Most campers in Seeler's study reported being forbidden by their parents or physicians to participate in certain sports. Almost all of them, however, admitted to participating in such activities without parental knowledge. The sports offered at the camp were the same as described

earlier. Although contact sports such as soccer or basketball were not encouraged, children who wanted to shoot baskets or kick a ball around were not forbidden to do so. Many boys were initially reluctant to enter into certain activities, using the excuse of supposed danger. In fact, the boys often simply did not understand the object or rules of the game, and were enthusiastic participants after having these explained. In concluding, she stated "Boys acquired skills that they can utilize in their daily lives, including sports which can be continued throughout the years. The self-confidence gained from such mastery was striking".

Boutaugh and Patterson have also reported on a camp specifically for hemophiliacs, this one sponsored by Hemophilia of Michigan, a chapter of the National Hemophilia Foundation.[24] Begun in 1969, the camp has had over eight years of experience and now accepts about 175 campers per year. The medical coverage was the same as in the previously described camp, with an emphasis on informality.

Activities available included fishing, swimming, lifesaving, canoeing, archery, basketball, badminton, tetherball, kickball, whiffle ball, ping-pong, and camping. Certain games, such as baseball, were modified to lower the risk of a hemorrhagic episode. Injuries and spontaneous bleeds did occur at the camp, but were treated promptly so that the camper could return to activity with minimum delay. The authors felt that athletic activity was a fun replacement for scheduled physical therapy, improving joint function and and muscle tone.

Children with inhibitors were accepted at the camp, and treated in accordance with the wishes of their hematologist at home. In the eight years of reported experiences no severe emergencies had occurred, but four of 689 campers had been sent home early either for previously existing medical problems or hemorrhages that did not respond to therapy. Overall, the camp was felt to be a fine personal growth experience for the patients. Self-image and confidence improved, and as one mother said, "J. went away to camp a young boy and came home feeling ten feet tall."

J. P. Allain, in describing his boarding school for hemophiliacs in France, stated that particular attention had been devoted to sports.[25] In his view, sports were beneficial to hemophiliacs from several points of view. They helped develop musculature, clearly providing an effective spontaneous physiotherapy and increasing the stability of joints. Psychologically, they provided an expression of aggressive urges, relieving anxiety. The hemophiliac, he stated, becomes aggressive, unafraid and competitive.

At his school every type of physical activity was permitted until its actual danger was proven. Although he did not go into the specifics of the situation, even games occasionally requiring body contact were allowed. His students competed against normal children in riflery, table tennis and swimming. Interestingly, the anxiety preceding these events did not produce the well-documented increase in spontaneous bleeding that would have been

expected. Examinations, on the other hand, were constantly preceded by increased bleeding.

Dr. Allain is no longer with the boarding school, and the philosophy of the institution has changed so that the duration of the boys' stay is now much shorter.[26] The new director, Dr. F. Verroust, has continued the policy of encouraging athletic participation. All of the boys currently engage in swimming and table tennis. Bicycling is also quite popular, but it is discouraged in those patients in whom it has been shown to cause increased bleeding. Although alpine skiing is not recommended, many of their patients do participate in cross-country skiing. The boys enjoy kicking a soccer ball around, but are not allowed to play actual games. When this was allowed in the past, several boys developed hematomas and hemarthroses. Interestingly, Dr. Verroust mentions that compliance is especially difficult with the restrictions in soccer and cycling, two of France's most popular sports.

Komp and Adams, in an article directly addressing the issue of athletic participation for hemophiliacs, stated that, "a carefully planned athletics program can play an important role in the total care of a hemophiliac."[27] They suggested four different beneficial effects of such a program: 1)the rehabilitation of the severely crippled child through recreational therapy, 2)the development of motor co-ordination, leading to fewer bleeding episodes secondary to clumsiness, 3)the stabilization of healthy

joints through strengthened musculature and 4)the development of healthy familial relationships and a positive self-image. They felt that if participation was encouraged in the safer sports, with emphasis on the positive, there would be less adolescent rebellion over restrictions, while dangerous sports, such as hockey and football, would be less appealing. Also emphasized was the fact that supervision and participation by the father gave him an important role in the life of his son.

Komp and Adams felt that a sport for a boy with hemophilia should be competitive but not provide body contact. Particularly suggested were swimming, tennis, air-riflery, angling, archery, badminton, billiards, bowling, fencing, golf, softball, ping-pong and volley-ball. Participation in basketball and track and field events was equivocal, while football, soccer, gymnastics, skiing and horseback riding were discouraged.

Lazerson, another outspoken advocate of sports for hemophilic children, echoed the often-heard opinion that the clinical course of the disease seemed to be less severe when the child was in good physical condition.[28] He contended that the increased stability of well-muscled joints led to fewer hemorrhagic episodes. In his opinion swimming was the best sport for hemophiliacs, and he recommended that all these children learn to swim by the time they were a year old. He also encouraged bicycling, basketball, touch football and baseball, stressing the importance of precautions such as the mandatory use of

batting helmets. As an example he reported the successful season of a six year-old severe hemophiliac in a youth baseball league.

Lazerson encouraged those hemophiliacs who excelled at a non-contact sport to progress as far as possible--even to the professional level. "What we need," he stated, "is a re-education of the public to allow hemophilic kids to participate and lead normal lives."

Finally, at least three articles have recently appeared in the popular press describing the athletic participation, and indeed triumph, of hemophilic children. One such youngster is Brian Deemer, a twelve year old severe hemophiliac from Covina, California.[29] Deemer pitched his team to an 18 - 2 record and the championship of his youth baseball league. Sustaining only one game-related injury during the season, he played in every game.

Jeff Browning is another hemophiliac who excels in athletics.[30] Participating in a sport more traditionally acceptable for hemophiliacs, Browning swam his way to a major letter this season on his school's swimming team. The younger of two hemophilic brothers from Milford, Connecticut, he is a 16 year old junior at Foran High School in Woodmont, Connecticut.

Perhaps the most impressive story is that of David Lerch, a twelve year old tennis enthusiast from Richmond, Virginia.[31] Ranked No. 1 in the state for his age group, Lerch is a severe hemophiliac. He began about five and one half years

ago, and progressed from beginning classes to public parks and recreation association programs and, finally, to state and regional tournaments. In addition to his state ranking, Lerch is rated No. 2 in singles in the Middle Atlantic Lawn Tennis Association. He also played on the No. 1 doubles team.

Although David suffers occasional bleeding episodes secondary to sprained ankles, his parents believe that the overall physical effect of his participation has been positive. This is especially true of his joints, where his increased musculature has led to improved stability.

Counseled by physicians to rear David with limited physical activity, the Lerchs decided that they "weren't going to raise a kid like that." As Mr. Lerch stated, "that wasn't our way of doing things. We decided that we were going to let him grow up a normal kid."

III. MATERIALS AND METHODS

The purpose of this investigation was to explore several aspects of the role of athletics in the life of the hemophiliac. In particular, the goals of the study were:

1) To determine the recommendations of physicians involved in the care of hemophiliacs regarding their patients' participation in sports.

2) To determine the actual extent of athletic participation by boys with hemophilia.

3) To determine the relative safety of the various sports currently being played by hemophiliacs.

4) To explore the opinions and attitudes of adult hemophiliacs regarding the role of athletic participation in the life of the boy with hemophilia.

The study was comprised of four distinct components, the methodology of which will be described separately.

I. Questionnaire Survey of Physicians

The physicians contacted for this aspect of the study were those practicing within the state of Connecticut who had expressed an interest in the care of hemophiliacs by registering with the Connecticut Chapter of the National Hemophilia Foundation to receive their general mailings. The list was composed of seventeen hematologists, thirteen general pediatricians, six orthopedic surgeons, three family practitioners and a physiatrist. Two physical therapists were also included. The physicians were sent a list of 45 sports and games and asked to indicate their reaction to each as either "Recommended for Hemophiliacs",

"Recommended only with Special Precautions" or "Not Recommended for Hemophiliacs."

Of the 43 questionnaires mailed, twelve were returned, five by hematologists, three by pediatricians, two by orthopedic surgeons, one by a family physician and one by a physiatrist.

II. Interviews with Hemophiliacs

The hemophiliacs interviewed were those residing the state of Connecticut who responded to an initial invitation for volunteers issued by the Connecticut Chapter of the National Hemophilia Foundation in their monthly newsletter. Of the 113 individuals registered with the Connecticut Chapter as having either factor VIII or factor IX disease, 21 volunteered to participate. In an attempt to identify a possible population bias, the 92 hemophiliacs who did not respond were sent a direct mailing and asked to indicate on a stamped, addressed postcard their reason(s) for not participating. Several possible reasons for declining inclusion in the study were listed on the postcard, along with a second invitation for those who now desired to participate.

Thirty-six postcards were returned and eleven more hemophiliacs volunteered to be interviewed. The breakdown of the reasons given by the other 25 respondents for their declining to be in the study were as follows:

I don't participate in sports	9
I didn't have time available	6
This study isn't worthwhile	2
This is an invasion of my privacy	0

I don't like discussing hemophilia	1
I felt I wouldn't be helpful because of my age	11
I didn't know about the research	4
The instructions were not clear	3
I am too severe to consider	1

Of the total of 32 volunteers, three were not interviewed because of an age of less than six years. One other volunteer was excluded because he had moved from the state. All other volunteers were interviewed, regardless of age, physical condition or area of residence in the state. The hemophiliacs involved in this study received their medical care from a wide variety of sources, including three university medical centers, a large university-affiliated city hospital and several different community hospitals.

The interviews were conducted in the homes of the volunteers by a fourth-year medical student. The tone of the interviews was casual and relaxed, and efforts were made to establish a healthy rapport, especially with the younger hemophiliacs. The interviews were semi-structured, with basic biographical and medical data collected at the onset. The hemophiliacs were then given the same list of 45 sports and games which had been presented to the physicians as described above. For each sport, the hemophiliac was asked:

1) Whether he had ever played the sport
2) How frequently he played the sport
3) Whether he had ever had a bleeding episode as a result of playing the sport (and if so, how often)
4) If he modified the sport in any way because of his hemophilia.

In the case of younger hemophiliacs this aspect of the

interview was conducted with the parents out of the room in an attempt to increase the accuracy of the answers when dealing with forbidden activities. Finally, the hemophiliac and/or his parents were asked open-ended questions about their thoughts on the value or danger of athletics for hemophiliacs. The interviews often went beyond the narrow confines listed above, giving the interviewer candid insights into the family's experiences with the disease.

III. Questionnaire Survey of Hemophiliacs

The hemophiliacs sent this questionnaire were those who had attended the summer camp operated exclusively for hemophilic youngsters by Hemophilia of Michigan. The largest camp of its type in the country, it draws campers from throughout the eastern half of the country. The questionnaire was included in a mailing from Hemophilia of Michigan to its 163 campers of last year.

The questionnaire was quite similar to the interview format described above. Basic biographical and medical data were requested, and the camper was presented with the same list of sports and games. For each activity, he was asked to report the frequency of his participation, any bleeds associated with that participation and any precautions taken to make the game safer. Of the 163 questionnaires sent, 28 were returned.

IV. Questionnaire Survey of Camps which Accept Hemophilic Campers

There are currently eighteen summer camps affiliated

with the National Hemophilia Foundation which accept hemophilic campers. The directors of these camps were contacted by mail and asked to list those recreational activities available to their campers with hemophilia. They were also asked to report any modifications or precautions used to make the activities safer, and any accidents serious enough to cause hospitalization of a camper. Of the eighteen camps contacted, eleven responded.

IV. RESULTS

I. Characteristics of the Study Population

Fifty-six hemophiliacs were included in the study. Twenty-eight of these were interviewed personally, the remainder being the respondents to the questionnaire survey. The age distribution of the study population is shown in Figure 1.

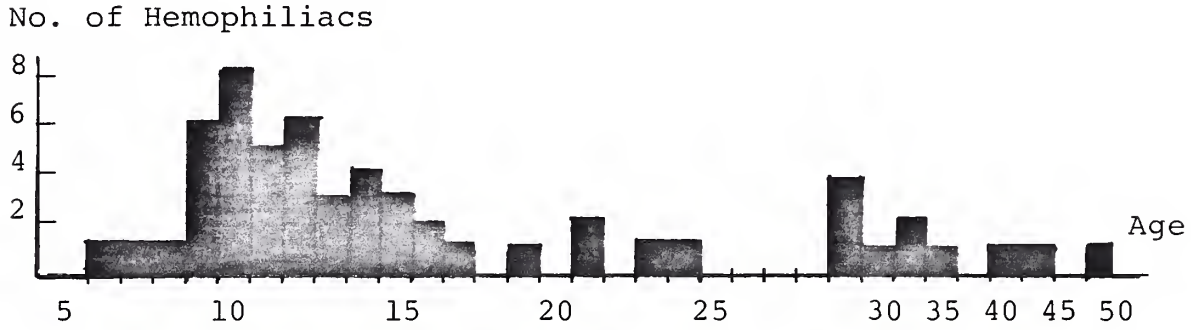

Figure 1

Forty-five of the hemophiliacs had classical hemophilia, while eleven suffered from Christmas disease. The breakdown with respect to severity of disease was as follows:

Severe	33
Moderate	18
Mild	4

Other questions asked and a tabulation of the results follow:

Do you have a brother with hemophilia? Yes 15 No 41

Do you have any other family history of hemophilia? Yes 24 No 30

Do you notice cycles of bleeding? Yes 46 No 9

Do you infuse yourself? Yes 30 No 23

How would you classify the majority of your bleeds? (From the choices given)

Spontaneous 21 Sports-related 10 Other Trauma 10

Who do you feel is the best judge of your ability to participate safely in a sport?

Self 26 Parent 7 Physician 5

II. Athletic Participation by Hemophiliacs

Table 1*

SPORT	N	PERCENTAGE OF HEMOPHILIACS WHO PARTICIPATED NEVER	RARELY	OCCASIONALLY	FREQUENTLY
Archery	52	35	35	15	15
Badminton	52	17	38	29	16
Baseball	53	25	11	38	26
Basketball	53	13	15	45	27
Bicycling	52	13	6	10	71
Billiards	52	25	29	21	25
Bowling	51	39	22	25	14
Camping	51	24	39	27	10
Canoeing	28	43	43	14	0
Croquet	52	44	33	15	8
Dancing	50	46	16	26	12
Fencing	52	100	0	0	0
Fishing	51	6	39	33	22
Football	53	25	15	28	32
Golf	52	58	23	15	4
Gymnastics	52	77	8	10	5
Handball	51	90	0	4	6
Hockey	51	80	12	8	0
Horseback	51	55	27	8	10
Hunting	51	76	12	10	2
Ice Skating	52	71	13	12	4
Kickball	40	17	10	38	35
Motorcycling	51	82	6	4	8
Ping-pong	53	26	32	21	21
Raquetball	52	86	6	2	6
Riflery/Skeet	52	62	19	13	6
Rollerskating	50	54	22	10	14
Rowing	51	53	29	8	10
Running	26	42	15	15	28
Sailing	52	77	13	4	6
SCUBA	52	90	6	2	2
Shuffleboard	52	60	31	7	2
Skiing	52	85	6	6	3
Sledding	51	10	16	61	13
Soccer	53	59	13	15	13
Softball	52	29	25	33	13
Squash	53	96	0	2	2
Swimming	52	0	10	29	61
Tennis	52	42	27	21	10
Volleyball	52	37	25	29	9
Water-skiing	52	90	10	0	0
Weightlifting	52	40	17	12	31
Wrestling	27	70	0	8	22

*See page 31.

*Notes on Table 1

For the purposes of this tabulation rare participation was defined as occurring less than twice per year. Occasional participation was defined as occurring between twice per year and twice per month, with frequent participation being more than twice per month.

In this study no distinction was made between formal and informal participation in a given sport. In general, organized participation in team sports was rare, and individual exceptions will be mentioned below. Modifications of the more violent contact sports were common. Touch football was played much more frequently than tackle, but almost all of those interviewed reported having played tackle at some time. Modified baseball, using either a whiffle or tennis ball in lieu of the hardball, was more common than the regulation game. Again, most of those interviewed had played with a hardball at some time. Hockey was generally modified so that the hemophiliac wore boots instead of skates, and the game was always informal in nature.

Because an important aspect of the study was to determine the actual pattern of athletic participation by hemophilic youngsters, the past participation in a given sport by an adult hemophiliac was included in the data.

Table 2

SPORT	PARTICIPATION INDEX*
Swimming	251
Bicycling	339
Kickball	191
Basketball	186
Fishing	177
Sledding	177
Football	167
Baseball	165
Billiards	146
Badminton	144
Ping-pong	137
Weightlifting	134
Softball	130
Running	129
Bowling	124
Camping	123
Archery	110
Volleyball	110
Dancing	104
Tennis	99
Croquet	87
Rollerskating	84
Soccer	82
Wrestling	82
Rowing	75
Horseback	73
Canoeing	71
Golf	65
Riflery/Skeet	63
Shuffleboard	51
Ice Skating	49
Gymnastics	43
Sailing	39
Hunting	38
Motorcycling	38
Hockey	28
Skiing	27
Racquetball	27
Handball	26
SCUBA	16
Squash	10
Water-skiing	10
Fencing	0

*PARTICIPATION INDEX=%Hemophiliacs partipating rarely in the sport + 2(%Hemophiliacs participating occasionally) + 3(%Hemophiliacs participating frequently)

III. Physician Recommendation and Camp Availability

Table 3

SPORT	PERCENTAGE OF PHYSICIANS (N=11) WHO RECOMMEND	RECOMMEND WITH PRECAUTIONS	DO NOT RECOMMEND	PERCENTAGE OF CAMPS OFFERING (N=1
Archery	91	9	-	64
Badminton	91	9	-	36
Baseball	18	45	36	36
Basketball	9	55	36	45
Bicycling	82	18	-	9
Billiards	100	-	-	-
Bowling	82	18	-	-
Camping	100	-	-	55
Canoeing	91	9	-	64
Croquet	100	-	-	-
Dancing	91	-	9	-
Fencing	36	-	55	-
Fishing	100	-	-	55
Football	-	27	73	-
Golf	100	-	-	18
Gymnastics	27	9	64	-
Handball	27	18	45	-
Hockey	9	9	82	-
Horseback	55	18	27	27
Hunting	64	27	9	-
Ice Skating	55	18	27	-
Kickball	36	18	45	18
Motorcycling	-	9	91	-
Ping-pong	100	-	-	18
Raquetball	36	27	36	-
Riflery/Skeet	82	9	9	9
Rollerskating	36	27	36	-
Rowing	64	27	9	36
Running	44	33	22	-
Sailing	82	9	9	27
SCUBA	36	18	45	-
Shuffleboard	100	-	-	-
Skiing	9	45	45	-
Sledding	20	30	50	-
Soccer	9	9	82	-
Softball	55	18	27	9
Squash	45	27	27	-
Swimming	100	-	-	100
Tennis	40	50	10	18
Volleyball	64	18	18	36
Waterskiing	9	18	73	-
Weightlifting	18	55	27	-
Wrestling	9	-	91	-

IV. Bleeding Associated with Athletic Participation*

Table 4

SPORT	N	PERCENTAGE EXPERIENCING BLEEDS NEVER	OCCASIONALLY	FREQUENTLY
Archery	12	83	17	-
Badminton	23	79	17	4
Baseball	31	45	39	16
Basketball	38	50	34	16
Bicycling	42	36	50	16
Billiards	23	96	4	-
Bowling	20	70	25	5
Camping	19	79	16	5
Canoeing	5	80	20	-
Croquet	10	40	60	-
Dancing	18	78	17	5
Fencing	0	-	-	-
Fishing	28	96	4	-
Football	31	13	58	29
Golf	10	80	20	-
Gymnastics	7	57	29	14
Handball	5	80	20	-
Hockey	4	50	25	25
Horseback	9	100	-	-
Hunting	6	83	17	-
Ice Skating	9	33	45	22
Kickball	26	42	42	16
Motorcycling	6	83	17	-
Ping-pong	20	100	-	-
Raquetball	4	75	-	25
Riflery/Skeet	10	90	10	-
Rollerskating	12	67	33	-
Rowing	9	100	-	-
Running	11	64	27	9
Sailing	5	80	20	-
SCUBA	2	100	-	-
Shuffleboard	5	100	-	-
Skiing	4	75	25	-
Sledding	38	61	29	10
Soccer	14	43	21	36
Softball	24	46	54	-
Squash	2	-	50	50
Swimming	46	87	13	-
Tennis	16	56	38	6
Volleyball	18	44	56	-
Waterskiing	0	-	-	-
Weightlifting	21	57	38	5
Wrestling	8	50	50	-

*See page 36

Table 5**

SPORT	BLEEDING INDEX
Squash*	150
Football	106
Soccer	93
Ice Skating*	89
Bicycling	78
Hockey*	75
Kickball	74
Baseball	71
Basketball	66
Croquet	60
Gymnastics*	57
Volleyball	56
Softball	54
Tennis	50
Raquetball*	50
Wrestling*	50
Sledding	49
Weightlifting	48
Running	45
Bowling	35
Rollerskating	33
Dancing	27
Camping	26
Badminton	25
Skiing*	25
Sailing*	20
Golf	20
Handball*	20
Canoeing*	20
Archery	17
Motorcycling*	17
Hunting*	17
Swimming	13
Riflery/Skeet	10
Billiards	4
Fishing	4
Rowing*	0
SCUBA*	0
Horseback*	0
Waterskiing*	0
Ping-pong	0

*N=less than ten
**See page 36

*Note on Table 4

In this tabulation N=the number of hemophiliacs who participated in the sport more than twice per year. Bleeding associated with 20% or less of the participations in a sport was termed occasional; bleeding occurring with more than 20% of the participations was termed frequent.

**Note on Table 5

The Bleeding Index was calculated from the data in Table 4. Bleeding Index=%Hemophiliacs reporting occasional bleeds + 2(%Hemophiliacs reporting frequent bleeding). The single asterisks (*) mark those sports in which fewer than ten hemophiliacs participated at least twice a year.

V. Interviews

In the course of the interviews, many sentiments were voiced by the hemophiliacs and their parents concerning athletic participation. These opinions and reflections of personal experiences covered a wide range of topics and provided a candid glimpse into the life of the hemophiliac and his family. Those comments which relate more directly to the topic of athletic participation will be described first, followed by a brief discussion of some broader issues.

Without exception, those interviewed felt that sports and games could offer significant advantages to the life of the hemophiliac, particularly in terms of psychosocial adjustment. Most simply felt that athletic participation was an important aspect of a normal childhood. Those aspects of athletics which were mentioned as the most beneficial included the sense of accomplishment, the experience of competition, the concept of fair play, and the camaraderie of teamwork.

In discussing the advantages of sports participation, those interviewed emphasized the psychological dangers of over-protection. Two hemophiliacs reported having engaged in illegal street racing as a result of being forbidden to compete in more acceptable athletic pursuits. Several other older hemophiliacs related similarly rebellious behavior as a result of the restraints on their activities as children. It should be noted that very little of this "daredevil" behavior was reported by the younger hemophiliacs or their

parents.

An overwhelming majority of those interviewed stated that they actually experienced fewer bleeding episodes during times of increased physical activity. This was generally attributed to increased joint stabilization leading to fewer spontaneous bleeds and freak accidents. A frequently cited example was the marked decrease in hemorrhagic episodes experienced during the summer, a time of increased activity for the younger hemophiliacs. One hemophiliac reported passing through a phase of increased bleeding associated with the onset of increased physical activity, only to break through to a phase of markedly decreased bleeding after a few weeks. Some examples of an improved clinical course following increased physical activity are summarized in the cases below:

Case 1 A young adolescent was on crutches continuously for over a year because of recurrent knee hemmorrhages despite prophylaxis. He joined the swimming team and was walking without crutches within a month. For the next year, he did not experience a single bleed. In his words, "If there is one thing I would tell a hemophiliac, it would be to join the swimming team. It was the best thing that ever happened to me."

Case 2 A nine year-old with an inhibitor was going through an especially severe cycle of bleeding. His parents began taking him to the YMCA on a daily basis. In the words of his mother, "I'm not sure if it helped, but the cycle ended.

Case 3 A thirteen year-old moderate hemophiliac played organized soccer last year. He suffered no injuries and noticed a marked decrease in the frequency of bleeding episodes during the season. This year he began participating in organized lacrosse, which has led to increased bleeding. In his opinion, his enjoyment of the game outweighs this negative factor.

Case 4 A thirteen year-old severe factor IX hemophiliac had a cycle of especially bad ankle bleeds despite prophylaxis. Continuing the prophylaxis, he began playing organized soccer. By the end of

the season, he had no difficulties with the ankle.

Case 5 A twenty-nine year-old severe hemophiliac began swimming twice per week two years ago. Since that time he has reported a marked decrease in bleeding episodes. He married at about the same time as he began swimming, and is not sure which had the beneficial effect.

Case 6 A twenty-three year-old severe hemophiliac began swimming daily when he entered college. He noticed a definite decrease in the frequency of his bleeding as a result. Later he began running, but this increased his knee and ankle bleeds. He continues to swim daily and run about 15 miles per week, but he now infuses concentrate before running. As he states, "The idea that I can be in good shape is amazing to me."

Obviously, not all athletic activity is accompanied by an improved clinical course. The interviews, the tables presented above, and even the cases just cited demonstrate that certain sports and recreational activities do lead to increased bleeding. The offending sports tend to be those involving body contact or significant joint strain, but exact correlation is difficult. Two of the youngsters mentioned above improved while playing soccer; two other hemophiliacs interviewed had increased bleeding when they began swimming on a highly competitive level. Other hemophiliacs interviewed had increased bleeding with any increase in physical activity. Many of these were surprisingly active, stating that their improved general sense of well-being more than outweighed the problem of the increased bleeding. Clearly, this is an issue with no exact answers. As many of those interviewed remarked, "Each hemophiliac has to find his own limits."

Concerning the issue of competition, most of those interviewed felt that it was a healthy component of

of recreational activity, improving confidence and self-image. A few of the older hemophiliacs felt that athletic competition was not appropriate for a hemophiliac. They believed that in the heat of competition, the hemophiliac would overstep his usual limits of physical exertion.

Two other benefits of athletics were mentioned anecdotally by the adult hemophiliacs. The first was the belief, expressed by a 44 year-old, that he had been better able to tolerate the increased plasma volumes administered in the days of FFP therapy because of increased cardio-vascular reserve developed from athletics. A 33 year-old self-admitted "sports freak" mentioned that in the worst of times, sports were something to look forward to, something to keep his spirits high.

An area often discussed in the interviews was that of the role of the parents in guiding the hemophilic youngster in his physical activity. There was unanimous agreement that simply forbidding a child to participate in a given sport was not only useless, but counter-productive. Those adult hemophiliacs who had been restricted from participating reported either defiance of the restriction or rebellion. Conversely, those hemophiliacs whose parents had not simply forbidden a given activity, but rather discussed the advantages and possible dangers of the sport, seemed to accept their limitations and found more appropriate avenues of athletic participation. Continually stressed was the importance of truly open communication between the hemophiliac and his family

regarding the disease and the decisions involved in its management. The hemophiliacs all expressed a need to be included in the decisions regarding their care.

This point is also reflected in one of the most commonly expressed complaints with the medical profession. Too often the hemophiliacs and their parents felt that their opinions had not been well regarded by physicians. Although most of those interviewed were satisfied with their own physicians, all were able to relate "horror stories" of their treatment at the hands of an unknown physician or emergency room. Many expressed the need for increased knowledge of hemophilia within the medical profession. They felt that there was widespread ignorance of the disease and its management among many segments of the health care system.

Public school systems were also frequently criticized for their lack of understanding of the hemophiliac and his problems. These students were often excluded from all physical education programs, prohibited from taking recess breaks outside with the other children, and generally made to feel "different" from their classmates. The significance of this remark is enhanced by the fact that a hemophiliac's desire to be treated like anyone else was the most commonly expressed opinion in the entire survey.

VI. Survey of Summer Camps

Camp directors reported remarkably few problems as a result of hemophilic campers engaging in sports. Traditional camp activities such as swimming, hiking, rowing, and archery were generally stressed, and most camps discouraged team sports such as soccer or basketball. The emphasis on non-team sports seems to be wise in view of the fact that such sports entail considerably less risk of injury than team sports, as is shown in Table 2 above.

When team games were a part of the camping program, they were generally modified to reduce the risk of injury. Modifications were also employed to allow participation by all campers, regardless of their infirmity. A common example was the use of whiffle or tennis balls in camp baseball games. In addition, substitute runners were often used for those campers who could not negotiate the basepaths on their own. At least one camp included the further provision that any player who attempted to slide into base was automatically "out".

Although games such as basketball or soccer were not encouraged, boys were usually allowed to shoot baskets or kick a ball around if they so desired.

In the summer of 1979 only one serious injury was reported by the eleven camps which serve a total of several hundred campers. This occurred when a junior counselor fell several feet from a tree. Tree-climbing was not an approved activity at the camp, and this type

of accident would have resulted in serious injury to any child.

V. DISCUSSION

Athletic and recreational activity is an important aspect of the lives of most boys and young men in our society. While many may feel that it is indeed over-emphasized, there can be no doubt that our culture considers athletic participation an important part of a normal childhood.[31] Because of the danger of bleeding, hemophiliacs were discouraged by their parents and physicians from such participation for many years. Within the last twenty years it was recognized that many hemophiliacs were in fact over-protected, being forbidden to engage in even reasonable physical activity.[10,11] This over-protection was believed to lead to significant psychosocial problems, among them rebellion, risk-taking behavior, and a passive-dependent character structure.[10,11,12] Because of this several authors have recommended reasonable athletic and recreational activity as one part of the overall approach to healthy psychological development in these patients.[11,12,13,22,23,25,27,28] Other authors, writing from the perspective of the physiotherapeutic discipline, have emphasized the potential value of sports in the maintenance of healthy joints.[5,6,7] This study was an attempt to determine the current extent of athletic participation by hemophiliacs. The relationship between this participation and their clinical course was examined, as were their opinions on the value of athletics in the development of a healthy character structure.

Perhaps the most fundamental conclusion of this study is the fact that the pattern of athletic participation by hemophiliacs closely parallels that which would be expected of the normal population. While hemophiliacs do make some concessions to their disease, such as modifications of the more violent contact sports, these are actually rather rare. The high frequency of participation in swimming, formerly the only sport recommended for these patients, is another example of their attempts to adjust their recreational activity to their disease. Other than swimming, the sports in which patients most frequently engaged were bicycling, kickball, basketball, fishing, sledding, football and baseball. When one considers this information in light of the relative incidence of bleeding episodes associated with participation in such sports, it becomes apparent that the sports most commonly played are generally among the more dangerous.

Despite the higher association of bleeding with many of the commonly played sports, it is important to stress that an overwhelming majority of those interviewed believed that an increased level of activity was definitely associated with a decreased overall incidence of bleeding. While this has never been proved in a controlled study, this paper reports several specific cases which demonstrate this point. Thus, the actual significance of the current pattern of athletic participation by hemophiliacs is unclear. While their high level of physical activity is encouraging, it is somewhat unsettling that their choice of recreational

activities includes the more dangerous sports.

In exploring the factors which affect a hemophiliac's choice of recreational activity, it is clear that physician recommendation has little influence. In this study the sports most frequently played by the hemophiliacs tended to be discouraged by the physicians. Twenty-six of 38 young hemophiliacs described themselves as the best judge of their ability to participate safely in a sport; only five felt that their physician was more qualified in this regard.

Parental proscription is also clearly ineffective in influencing the hemophiliac's choice of activities. In this study, 20 of 26 young hemophiliacs admitted to participating in sports which were expressly forbidden by their parents. Many of the adult patients agreed, stating that restriction only leads to rebellion.

It appears that the most important factors in the hemophiliac's choice of sports are cultural and peer pressures. The sports he chooses to play are those that his friends play and his culture rewards. In the United States, these are largely the competitive team sports such as football, basketball, and baseball. As an example of a different cultural influence, in France it is soccer and cycling which are most popular. In both countries, these popular sports lead to conflict between the hemophiliac and his physicians.

Regardless of the culture, the physician must understand that the factors affecting the hemophiliac's choice of recreational activity are largely beyond his control.

The physician cannot expect his advice to be fully accepted by the young hemophiliac, and he must be aware that the child will participate in sports and games which the physician does not recommend. An important corollary of this is the fact that a hemophiliac may be reluctant to discuss a sports-related bleeding episode with his physician if he believes that the physician will use that discussion to criticize the participation.

While a hemophiliac cannot necessarily be prevented from a participating in a dangerous activity, there may be means by which a family can lessen the allure of such sports.

One approach to this situation is suggested from the lack of conflict over athletic participation seen in those families where the disease and its management were openly discussed with the hemophiliac. In these families the son was included in the decision-making process regarding his care. Many of the adult hemophiliacs felt that this form of open communication was essential to prevent rebellion.

Combined with this approach is the concept that the young hemophiliac should be directed toward the safer sports, rather than away from the more dangerous. If the child develops a true interest in a safe sport, the more dangerous will seem less appealing. The attraction of those sports now commonly played by hemophiliacs is their popularity in our society at large. If a child is introduced to a safer sport through his family, it can carry the same importance that our culture assigns to the more dangerous

games. This is not to say that the hemophiliac should be forced into any particular activity. Rather, if the child has a pleasant exposure to a safe sport while young, the other sports will hold less appeal. This approach has another benefit as well. In addition to introducing the young hemophiliac to an appropriate sport, it offers an excellent opportunity for strengthening the father-son relationship, traditionally a weak point in these patients' family dynamics. The sports best suited for this approach are the so-called "life sports" (i.e. swimming, tennis, golf, etc.) which have a lower incidence of associated bleeding episodes and which will afford the hemophiliac a lifetime of recreational activity. Furthermore, these sports offer a more realistic opportunity for formal competition than do the more dangerous team sports.

This approach is only a guide. Each hemophiliac and his family must decide for themselves which sports are appropriate. Also, it must be remembered that despite the most cautious approach, some bleeding episodes will occur as a result of athletic participation. The advantages of this participation, however, are many and far-reaching.

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