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Adamantinoma Of Tibia

Volume XIV | Friday, October 9, 1942 | Number 2

STAFF MEETING BULLETIN
HOSPITALS OF THE . . .
UNIVERSITY OF MINNESOTA

Volume XIV | Friday, October 9, 1942 | Number 2

INDEX

		PAGE
I.	LAST WEEK	22
II.	MEETINGS	
	1. SEMINAR IN PATHOLOGY	22
	2. PHYSIOLOGY-PHARMACOLOGY SEMINAR	22
	3. CLARENCE MARTIN JACKSON LECTURE	22
III.	ANNOUNCEMENT	22
IV.	ADAMANTINOMA OF TIBIA	
	. . . . J. C. McCartney, G. R. Dunn and E. T. Evans	23 - 29
V.	GOSSIP	30

Published for the General Staff Meeting each week
during the school year, October to June, inclusive.

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Alumni and Friends

William A. O'Brien, M.D.

I. LAST WEEK

Date: October 2, 1942

Place: Recreation Room
Powell Hall

Time: 12:15 to 1:15 p.m.

Program: "Hospitals Report - 1940-1942"
R. M. Amberg

Movie - "Sulfonamide Therapy"

Discussion
F. E. Schmidt

Present: 87

Gertrude Gunn,
Record Librarian

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II. MEETINGS

1. SEMINAR IN PATHOLOGY

Etiology of Hypertension - E. T. Bell. Monday, October 12, 1942, 12:30 p.m., 104 Anatomy Bldg.

- - -

2. PHYSIOLOGY-PHARMACOLOGY SEMINAR

The Physiology-Pharmacology Seminar will meet in Room 214, Millard Hall on Tuesday, Oct. 13th at 12:30 P.M.

R. N. Beiter will speak on "Chemotherapy of Experimental Malaria Infection."

Maurice B. Visscher.

- - -

3. CLARENCE MARTIN JACKSON LECTURE

Wednesday, October 21 at 8:15 p.m. in Museum of Natural History.

Dr. Thomas Francis, Jr. will speak on "An Interpretation of Current Studies in the Control of Epidemic Influenza."

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III. ANNOUNCEMENT

BULLETIN - General Staff Meetings

Each week a bulletin will be issued for the general staff meeting of the University of Minnesota Hospitals. A copy is given to each person who attends. In addition they are sent by mail to interested persons.

In order to defray part of the expense of mailing the bulletin a charge of $2. for the 1942-1943 series will be made. Each bulletin will be punched for binding. Covers will be included in the above subscription rate. As there is a shortage of metal the usual clasp cannot be obtained. In its place a string will be used to bind the series together.

Many distinguished names have been found on our subscription list during the past years. An open invitation is extended to any interested person to subscribe for the series. Address all communications to Miss Gertrude Gunn, Record Librarian, University of Minnesota Hospitals, Minneapolis, Minnesota. Please enclose check or money order with subscription.

IV. ADAMANTINOMA OF TIBIA

J. S. McCartney
G. R. Dunn
E. T. Evans

Since Hebbel published his report of a case of adamantinoma of the tibia in 1940, but two additional instances of this type of tumor have been reported. These two cases were reported by Dockerty and Meyerding. The purpose of this report is to record the outcome of Hebbel's case and to report 2 new instances.

That tumors resembling the enamel organ should develop in the maxilla and mandible or in adjacent parts of the head or even in the ovary is hardly to be wondered at, but the occurrence of such tumors in other parts of the body is surprising, particularly when the site of occurrence is apparently limited to one bone, the tibia. With the publication by Fischer of the first instance of this type of tumor in the tibia, speculation as to its origin and etiology began and has continued with but little, if any, clarification. As Robinson points out, the tibia is the only site in which adamantinoma occurs where it may be necessary to postulate an epithelial rest. Epithelial rests in the jaws are common. While perhaps such a rest of ameloblasts in the tibia to the exclusion of all other bones might be considered "mythical" (Dockerty and Meyerding), nevertheless it can hardly be dismissed as a possibility. Trauma with deep ruptures of the epithelium and laceration of the periosteum with implantations of epithelium and "thwarted repair" as suggested by Ryrie is perhaps hardly more tenable than the embryonic rest theory. Ryrie contends that the relations of the tibia to the overlying skin make it the site where such deep ruptures and implantations can most readily occur. This is hardly valid, since there are surely portions of other bones which are also closely related to the overlying skin, and which are not infrequently traumatized, e.g., the radius and ulna, and yet adamantinomas do not occur in these bones. However, one case of primary carcinoma of the ulna has been reported by Maier. Her patient, according to Kronlein, was alive and well six years and nine months after operation. Further the fate of implanted epithelium is ordinarily complete absorption or formation of an epidermoid cyst. The possibility of tibial adamantinoma being a unilateral development of a teratoma is a possibility, although the extremities are uncommon sites for teratomas, and why should the tibia alone be involved. Still a number of authors have noted the similarity of the microscopic structure of tibial adamantinoma to that of mixed tumors of the salivary glands. An endothelial origin has been suggested and Richter recognizing the similarity of his tumor to that of Fischer's, but believing in the endothelial origin, preferred to call his tumor "adamantinoma-like." Resemblance of tibial adamantinoma to basal or squamous cell carcinoma has been noted by several writers, but it is clear that tibial adamantinoma cannot be a secondary involvement of bone from an overlying skin carcinoma. When there has been involvement of the skin with tibial adamantinoma it has occurred long after the tibial tumor was first noted. The overlying skin has usually been said to have been intact.

While it must be admitted that 14 of the known cases of adamantinoma of the tibia had given a history of trauma it is difficult to believe that the trauma was the cause of the tumor. In many instances the time from the trauma to the appearance of the tumor seems to have been too long to admit of any connection. On the other hand in some there have been continuous symptoms from the injury until the appearance of the tumor. If trauma were the only factor tibial adamantinomas would probably be frequent, instead of being curiosities. As yet no entirely satisfactory theory for the development of adamantinomas in the tibia (to the exclusion of all other bones) has been advanced. If it be admitted that simulation of the enamel organ is simply one of the ways in which ectoderm may differentiate, then primary tibial squamous carcinomas belong in the same category. By the same sign, Maier's case may be added to the

total and thus the tibia is not the only site where primary epithelial tumors of bone may develop (mastoid?).

Attention should be called to the fact that of the nineteen known instances of tibial adamantinoma, fifteen were in individuals under 40 years of age and indeed ten of them were under 30 years of age. The ages ranged from 12 to 57 years, 12 were males and 7 females. This may suggest that this age distribution is evidence in favor of the embryonic rest theory. It is surely earlier in life than the usual time of appearance of squamous cell carcinomas of the skin, but it can hardly be argued that the earlier appearance favors the differentiation toward enamel organ rather than squamous epithelium. Indeed in a number of cases squamous e;ithelium and epithelial pearls have been noted.

Case Reports

Hebbel's patient was a girl, 14 years of age, who had had difficulty since December 1935. The symptoms were pain, tumor of the right leg and difficulty in walking. The lesion was curetted in April 1936 with disappearance of symptoms for six months, when they recurred. Curettage was repeated in July 1937 and it was followed by relief. Again symptoms recurred and she was admitted to the University Hospitals in July 1938. Here after diagnostic curettage a low thigh amputation was done. From the biopsy material one of us (JSMc) made the erroneous diagnosis of sarcoma. As can be seen from Hebbel's photomicrographs such a diagnosis is hardly to be wondered at. Within a month after operation the patient noted a small hard nodule growing from the upper medial portion of the thigh. This nodule progressively enlarged so that at the time of readmission, March 20, 1941, it was the size of a large orange. She said that she felt well, but had lost 11 pounds in weight in three weeks "due to a cold." Cough has been present for a week. There was no sputum. Also for the past week she had had exertional dyspnea. Physical examination at this time revealed dullness to flatness at the right base posteriorly and enteriorly beginning at the 6th to 7th dorsal spines. Breath sounds, vocal and tactile fremitus were decreased at the right base posteriorly. Breath sounds were decreased posteriorly over the 4th to 7th spines. There was questionable dullness in this area. The right thigh showed a mass the size of a grapefruit protruding from the upper inner aspect of the thigh. This mass was movable but seemed to be attached to the bone. It was firn, not tender and questionably cystic. No enlarged lymph nodes were found. X-ray examination revealed multiple metastases in both lung fields, and a bilateral pleural effusion. About March 31, 1941 and after discharge she developed a Type VII pneumonia. A follow-up revealed that she had died January 23, 1942 after gradually failing in strength. For the last month she had constant severe pain in the chest. An X-ray examination made 3 days before death showed extensive involvement of both lungs. An autopsy was not done. While it might be argued that there is no absolute proof the pulmonary lesions were metastases from the tibial tumor, it seems a safe conclusion. No other instance of metastasis and death from tibial adamantinoma has been reported and this case is of interest from that standpoint alone. This patient lived just over six years from the onset of symptoms. The gross and microscopic features of this case have been well described and illustrated by Hebbel.

Case 2.

This patient, a male 32 years of age, was first seen on May 10, 1940, complaining of a tumor of the left leg. According to the history obtained at that time he had bruised his leg first in August, 1931 and again in July 1932. Because of disability he stopped work on 2 or 3 occasions and during the fall of 1932 the left tibia was operated upon for a "peculiar cyst." At a second operation in 1933 a diagnosis of carcinoma was made. X-ray treatment was given. He was in another hospital in 1934 because of his tumor. From this time he got along well until 1940 when he was in still another hospital. The leg was operated upon in February 1940

and the diagnosis of adamantinoma made. He was treated with X-ray. When first seen by one of us (ETE) he was having no pain but showed a skin metastasis or direct extension, a long old operative scar, and an ulcer over the tumor. A node the size of a hazel nut was present in the left inguinal region and a smaller one in the right. X-ray showed almost complete destruction of the middle and lower thirds of the left tibia, and a pathological fracture was present. The chest was clear. A cast was applied. On June 7, 1940 the enlarged node was removed from the left inguinal region. It showed only simple adenitis. Biopsy of the leg tumor was done on July 12, 1940 and revealed Squamous Cell Carcinoma. A Kirk type of amputation was finally performed on September 3, 1940.

The amputated extremity showed an ulcer about $2\frac{1}{2}$ cm. in diameter on the anterior aspect of the tibia over the lower third. When the extremity was split lengthwise a tumor rather sharply outlined and somewhat lobulated and cystic was found continuous with the skin ulcer and extending through the muscles and into the tibia. The tumor had over-all measurements of 15x6x6 cm. Microscopic examination of this tumor showed why a diagnosis of squamous cell carcinoma was made at the time of biopsy in July 1940. Many areas were composed of cords and masses of squamous epithelium in a compact fibrous tissue. In these areas there was nothing to suggest adamantinoma. In other areas the structure was that of spindle cell sarcoma. In some parts there was a reticulated arrangement, similar to that seen in the central part of a typical adamantinoma. However, the peripheral deeply staining mass of epithelial cells which go to make up the rest of the enamel structure were lacking. It was only after comparison with the findings in the various parts of the sections with those of other adamantinomas that a diagnosis of adamantinoma appeared justified. Hebbel and others have beautifully shown the great variations in the microscopic appearances in tibial adamantinomas.

Case 3

This man, 39 years of age, was injured August 15, 1940 by a piece of pulp wood 8 feet in length falling and striking his right leg over the shin bone. He had considerable pain and that night his leg was swollen and quite painful. The leg became black and blue from the knee to the ankle. He did not work for about two and one half days. The swelling gradually disappeared, but a small lump over the shin persisted. In about three weeks all swelling had gone, but the lump was always a little sore and sensitive, especially when he would bump it. Otherwise it was rather numb. He continued to work until April 3, 1942. He first consulted a physician in August, 1941 but not because of the leg tumor. The physician said a tumor might be developing. The patient thinks the lump has enlarged slightly, and since September, 1941 has grown more rapidly and become quite painful. A tumor in the bone was first demonstrated by X-ray on April 12, 1942. When first seen by one of us (GRD) on May 15th the patient's leg was so painful that he could hardly work.

Physical examination at this time showed marked gingivitis and pyorrhea. The lower extremities showed a mass on the anterior and medial aspect of the right leg at about the region of the junction of the middle and lower thirds of the tibia. This mass was firm, rather tender, localized and about the size of a hen's egg. The tenderness was chiefly of the lateral and inferior portion of the mass. Slight tenderness over the lower portion of the tibia was present. There was no redness, edema or other evidence of circulatory disturbance. Slight varicosities of the other leg were present. Otherwise the physical examination was negative. Laboratory studies showed hemoglobin 104%, red blood cells 5,260,000 and white blood cells 16,200. The differential count was 66% neutrophiles, 27% lymphocytes, 2% band forms, 4% eosinophiles and 1% monocytes. Blood chemistry studies showed calcium and phosphorous levels within normal limits. The urine was normal. The report of the

X-ray examination was as follows: "Antero-posterior and lateral films were made of the right tibia and fibula. These show a large multilocular area of bony destruction in the middle third. The shaft of the tibia is expanded in this region and there is evidence of some periosteal proliferation. The proximal third of the tibia is normal. X-rays of other bones normal. Conclusions: The lesion in the shaft of the tibia described above is atypical. I believe this represents either a fibro-cystic disease or possibly a giant cell tumor. The weight of evidence is in favor of the former. I also believe that this lesion, while it has a malignant appearance, is benign. I suggest that the possibility of a hyperparathyroidism be investigated."

On May 30, 1942 biopsy of the tumor was done and a microscopic diagnosis of adamantinoma made by one of us (JSM). Immediately 13 cm. of the tibia was removed and a bone graft from the opposite tibia inserted. Postoperatively convalescence was complicated by a bilateral phlebitis. The patient has now fully recovered.

The excised portion of tibia measured 13 cm. in length, and consisted of the entire thickness of the shaft. There was a defect in the surface through which the biopsy specimen was removed. (Before biopsy the tumor was entirely within the bone, and covered on all surfaces by bone.) On splitting the tibia lengthwise a tumor measuring 9x3x3 cm. was found. It had a mottled appearance, in part being pale, in part red. It was lobulated and partially cystic. Above and below the tumor seemed to blend with the marrow, no sharp line of demarkation being present. Microscopic examination: The tumor was composed of structures apparently identical with those seen in adamantinoma of the jaw, namely masses and cords of intensely staining cells peripherally and loose reticulated centers. These cords of cells were scattered through fairly compact connective tissue. In a few areas cornified masses of epithelial cells were present.

As has been mentioned by several writers the illustrations of the x-ray, gross and microscopic appearances of tibial adamantinoma are practically interchangeable for all cases. The X-ray reveals a multilocular destruction of the bone with some expansion. The gross specimen shows a somewhat lobulated tumor involving cortex and medulla. The tumor may spread out beneath or perforate the periosteum. The overlying soft tissues may be pushed aside or invaded. As in our second case the skin may be invaded and ulcerated. The microscopic picture is extremely variable and in the absence of structures resembling typical adamantinoma an erroneous diagnosis may be made. In addition to the usual appearances of adamantinoma areas simulating sarcoma or squamous cell carcinoma may be present.

The usual history of the course of tibial adamantinoma is one of repeated recurrences and eventual amputation, although a few have apparently been cured by resection. In most of the reported cases the period of follow-up has been too short to claim a cure. One of Wolfort and Sloane's patients had no further recurrence 47 months after the first operation, and Maier's patient with the ulnar carcinoma was alive and well after six years and nine months. It is apparent from a study of the reported cases that the course of tibial adamantinoma is prolonged.

Summary

1. Follow-up and report of death from a previously reported case of tibial adamantinoma is given. Two new cases are reported.

2. The bulk of these tumors occur in the first half of life.

3. The etiology and pathogenesis of tibial adamantinoma is obscure.

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SUMMARY OF CASES OF ADAMANTINOMA OF THE TIBIA

Case	Year	Author	Age, Yrs.	Sex	Site	Antecedent Trauma	Latent Period	Character and Duration of Symptoms	Initial Therapy	Subsequent Course
1	1913	Fischer	37	♂	Junction lower and middle thirds, left	Contusion	5 mo.	Pain, swelling, 5 mo.	Resection: bone graft	No recurrence 8 mo. later
2	1930	Richter	12	♂	Middle third, left	Fall	?	Swelling, 8 mo.	Amputation	Uneventful.
3	1931	Baker and Hawksley	46	♂	Lower third, left	Contusion	8½ mo.	Pain, swelling, 6 wk.	Resection; bone graft	No recurrence 2 mo. later.
4	1932	Ryrie	52	♂	Middle third	Contusion	8 yr.	Swelling, 8 yr.	Curettage	Recurrence; amputation 6 mo. later.
5	1933	Petrov and Glasunow	22	♂	Middle third, left	None	None	Swelling, 1 yr.	Resection; bone graft	No recurrence 1 yr. later.
6	1934	Holden and Gray	36	♀	Lower third, left	Abrasion, contusion of ankle	None	Pain, 2 yr.	Excision	Recurrence after 2 yr.; reexcision; roentgen therapy of high voltage.
7	1937	Bishop	22	♂	Upper third, right	Fracture	8 wk.	Swelling, 3 yr.	Curettage, chips, graft	Recurrence; amputation after 14 mo.; 4 yr. after fracture.
8	1938	Renbock and Barber	24	♀	Lower third, left	Sprain of ankle	None	Pain, swelling, 18 mo.	Wide excision	No recurrence 8 mo. later
9	1938	Oberling, Vermes & Chevereau	51	♂	Middle third	?	?	?	Excision	Recurrence; amputation 7 mo. later.
10	1938	Dunne	32	♂	Junction upper and middle thirds, left	Contusion, abrasion	9 mo.	Swelling, 4 yr.; pain later months	Roentgen therapy of high voltage	Progression; amputation 9 mo. later.

SUMMARY OF CASES OF ADAMANTINOMA OF THE TIBIA (Cont.)

Case	Year	Author	Age, Yrs.	Sex	Site	Antecedent Trauma	Latent Period	Character and Duration of Symptoms	Initial Therapy	Subsequent Course
11	1938	Wolfort and Sloane	57	♀	Junction middle and lower thirds, left	Contusion	None	Swelling, 19 mo.; pain, 4 mo.	Excision	Recurrence in 16 mo.; resection; no further recurrence 47 mo. after 1st operation.
12	1938	Wolfort and Sloane	18	♀	Lower half, right	Abrasion	3-2/3 yrs.	Pain, 4 mo.	Curettage, radium	Recurrence; resection 13 mo. later; recurrence again; amputation -12 mo.
13	1939	Thomas	19	♀	Upper right	None	None	Swelling, 7yr. pathologic fracture	Curettage, graft	Recurrence; amputation 8 mo. later.
14	1939	Rankin	25	♂	Lower left	Puncture wound	1 yr.	Pain, swelling, 1½ yr.	Curettage.	Recurrence; excision 15 mo. later; no recurrence 2 yr. after first operation.
15	1939	Hobbel	14	♀	Lower right	None	None	Pain, swelling, 4 mo.	Curettage.	Recurrence; curettage 15 mo. later; again recurrence, amputation 2½ yr. after onset.
16	1941	Docherty and Meyerding	24	♀	Left middle	?	None	Recurrent pain 8 yr.; swelling, 4 yr.	Excision	Recurrence; amputation 15 mo. later.
17	1941	Docherty and Meyerding	27	♂	Left middle	Abrasion	6 yr.	Recurrent fractures; pain; tumor	Repeated excision; roentgen therapy	Arrested (?) 16 mo.
18	1942	McCartney	32	♂	Left lower	Bruised	1 yr ?	9 yrs.	Multiple excision	Amputation 2 yrs.
19	1942	McCartney	39	♂	Right middle	Bruised	?	Pain, tumor	Resection	Resection 3 mos.

V. GOSSIP

This column is devoted to news items of interest to the staff and those who subscribe to the bulletin. Much that is contained here may be of interest to all. At other times it will concern just a few and will replace a letter which might be sent to cover the same news. Please forgive if at times most of the items seem to be of interest to the "other fellow"....Paul Oberg, new chairman of the School of Music, reports that Dr. Herald L. Lamb, who graduated from the University of Minnesota in 1902 is a member of the University of Minnesota Symphony Orchestra. He plays in the violin section. Each week he makes the trip from Little Falls for the regular practice period. Like many other physicians he finds that a hobby pays rich dividends. In Duluth the doctors are well organized in music. At the reunion meeting of the interns of St. Mary's Hospital this summer the doctors' orchestra played for breakfast. It was an unusual experience to hear light opera with your orange juice and ham. The group was under the leadership of Dr. William J. Ryan. Dr. Ryan, a brother of Drs. Mark and John Ryan of St. Paul, is said to be the only member of the Symphony Orchestra who walked out on a conductor and still holds his job. According to the Reader's Digest, Dr. Ryan deserted his place to take care of a woman in labor....Dr. George Hauser, our "new" coach, is having a successful season. It is true the scoreboards showed that the Seahawks kicked a goal, but otherwise Minnesota received the credit for a whale of a performance last Saturday. George has been the students' friend for a long time. They have confidence in him and the remarkable physical condition of the squad is a direct tribute to his insight and planning. Few squads get better care when accidents occur. Employing a physician on the squad for advice is ideal. In the practice season the players are fed three meals (also on the road). At home they get one meal after practice. This is the usual good meal which has been served in the past. Football players need between 8,000 and 9,000 calories a day to maintain weight and physical well-being. Those who have witnessed the squad in action at the table marvel that so much food can be put away in such a short time. In addition to his care of the students George and his staff are turning out one of the best drilled teams that Minnesota has produced in many a day...The alumni are coming back for their homecoming as usual this year. The dates are October 21-24. On Wednesday, October 21, Dr. Francis will give the Clarence Martin Jackson lecture. On Thursday and Friday, October 22 and 23, there will be a clinical program. On Saturday morning the group will attend the clinical-surgical-radiological conference and in the afternoon witness the game between Michigan and Minnesota. This event will mark the 50th anniversary of football rivalry between these two schools. Fifty years is a long time to carry on an athletic tradition, but our arrangement with Wisconsin is older than this. There will be a reunion of the class which graduated 20 years ago in 1922. The group will consist of the December class of 1921 and the winter and spring classes of 1922. These students started together and although they did not finish together they were the class of 1922...Staff meeting last week got off to a good start. The number in attendance was in keeping with the decreased number on the staff. We anticipate a weekly attendance of between 90 and 100 this year and see no reason why the program cannot be as effective as ever....The senior students were given a course in war medicine this summer. More than 30 lectures comprised the series. During the month of September they read selected references in anticipation of a test to be given during the first week of school. This test revealed that as far as the clinical problems ahead are concerned most of our students are well informed. It appears that they are taking extra interest in the subject. The freshman students are finding their course in "first aid" of greater importance than ever. The new textbook in advanced first aid by Cole and associates from the University of Illinois makes interesting reading. A city is now believed to be well protected if 5% of its citizens have successfully completed a course in first aid. In the midwestern section St. Paul and Houston, Texas are the only cities which have qualified to date. St. Paul in its own way and without too much publicity has made many interesting health records.....