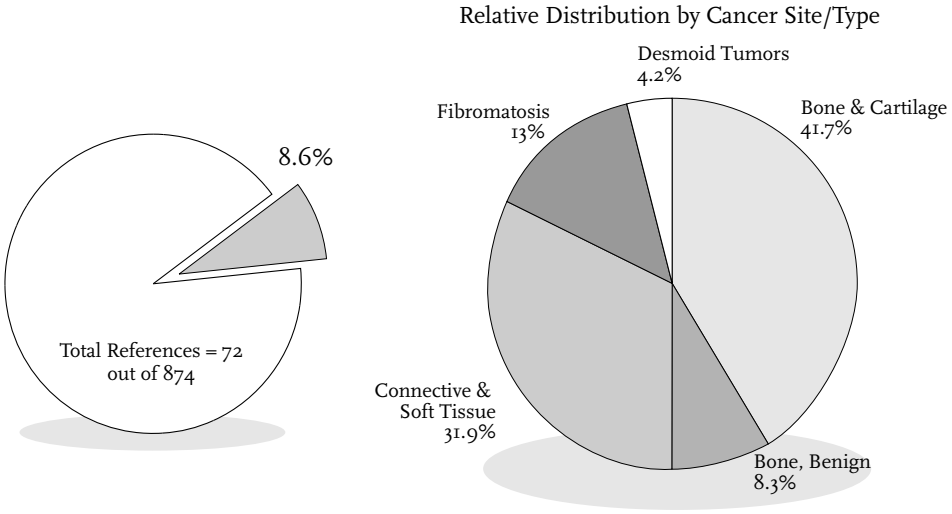
4. Remission of Neoplasms of Bone, Connective Tissue, and Soft Tissue

Remission of Neoplasms of Bone, Connective Tissue and Soft Tissue



References in Chapter Four = 72
References in Part One = 874

Cancers of the bones and joints and soft tissue (including heart) account for 0.8% of the cases of cancer reported by participating tumor registries to the SEER (Surveillance, Epidemiology, and End Results) Program between 1983 to 1987. Cancers of the bones and joints comprised 0.2% and soft tissue cancers (including heart neoplasms) comprised 0.6% of all cancers reported during those years. The relative five-year survival rates (%) for the years 1981-1986 for musculoskeletal and soft tissue cancers are 53.4% for bone and joint cancers, and 60% for soft tissue cancers. The average annual age-adjusted mortality rates for 1983-1987 for neoplasms of the bone and joints was 0.4 per 100,000 population in the United States and the mortality rate for soft tissue neoplasms (including heart) was 1.1 per 100,000 population (Cancer Statistics Review 1973-1987, published by the National Cancer Institute).

There are 72 references in Chapter 4. Twenty-eight references are annotated with summaries. Some annotated references also contain 1 or more case reports. Forty-four supplemental references are cited to provide additional research information. The full text of 27 case reports is presented.

The chapter contents are summarized in Table One. A comparative analysis of cases reported in previous literature reviews is presented in Table Two.

Table One: References and Case Reports in Chapter Four†

Tumor Site/ Type	References (number)	Cases (number)	Cases (%)
Bone (malignant)	30	6	2.3%
Bone (benign)	63	6	2.3%
Connective Tissue (malignant)	23	7	2.7%
Connective Tissue (benign)	3	3	1.2%
Fibromatosis	10	5	1.9%
Totals	72	27	10.4%

† Total number of case reports in Part One is 258.

Table Two: Comparison Between Other Major Literature Reviews of Spontaneous Regression of Neoplasms of Bone, Connective Tissue and Soft Tissue

Tumor Site/ Type	Rohdenburg (1918) (N =185)	Fauvet (1960) (N=192)	Boyd (1966) (N=97)	Everson (1966) (N=182)	Challis (1990) (N=505)
Bone (malignant)	5	3	2	8	3
Connective Tissue (malignant)	18	19	4	11	3
Totals	23	22	6	19	6

Malignant Neoplasms of Bone and Cartilage

Diet in Cancer

First Paper: Full Text of Nine Cases

Cutter E

Albany Medical Annals 8: July 1887; 218-230

Extracted Summary

This paper is intended to be practical, giving histories of some cases where there were special diets adopted which seemed to be beneficial. It is offered as a contribution to medical knowledge to point out the way in which the writer thinks that organic disease should be approached, that is, through the function of nutrition; to show that alimentation is an agent of tremendous power, and to impress the idea that diseased tissues are sometimes amenable to food influences even in apparently desperate instances. In a second paper the theoretical side of the question will be considered.

Selected Case Report

Case 2. Diet of Bread; Infusion of "Water Dock"; Cure. "Dr. W. H. Thayer, in a letter to me, says: 'I have obtained from Dr. Twitchell all the particulars of the case of treatment of osteosarcoma which he could give me; and as his memory is so accurate, I suppose he has not forgotten anything of importance connected with it. You know the doctor never takes notes.'

"A man about forty years of age consulted Dr. Twitchell in relation to a tumor on his scapula, as large as a pint bowl. It was evidently osteosarcoma, had its usual crackling feel, and resembled very closely one in the same position which Dr. Twitchell had seen a short time previously, and for which he had removed the whole upper extremity, even scapula and clavicle. In that case the wound healed, but the man died a year or two afterwards with carcinoma of some internal organ.

"When the second case applied for advice, Dr. Twitchell declined an operation, and the man returned home to Vermont. Soon afterwards he heard of somebody in New York who could cure him, and, applying to this person for advice, received the following: He was to take from the brook which ran through his native farm a plant which grew there (the adviser did not say what it would be), and use a weak infusion of it for his only drink every day until the tumor had disappeared. His diet, besides this, was to consist of bread alone. This advice was strictly followed; the plant he used was 'water dock.'

"Dr. Twitchell happened to see the man two years afterwards, when he was still following this course. He found the tumor had nearly disappeared, there being apparently only a trifling thickening of the skin."

Primary Malignant Tumors of the Long Bones

End Results in 170 Operable Cases

Coley WB; Coley BL

Archives of Surgery 14: 1927; 64-141

Extracted Summary

The prognosis of osteogenic sarcoma of the long bones, while far from satisfactory, is by no means as hopeless as is generally believed by physicians. The prognosis depends largely on an early diagnosis and the exercise of most careful judgment in selecting the method of treatment for the individual cases; this should be based on a wide experience with a great variety of clinical and pathologic types of bone sarcoma.

Amputation alone offers little hope of a permanent cure in any of these types. Primary treatment with radium or roentgen rays, even if pushed to the utmost limits of safety, while often

causing very marked improvement has thus far failed to effect a permanent cure in any case in which the diagnosis has been unquestionably established by clinical and pathologic evidence. The mixed toxins of erysipelas and Bacillus prodigiosus alone have effected a cure in certain number of cases; but all of these cases have been of the round cell or spindle cell type, characterized by little production of new bone. The mixed toxins and radium combined have likewise resulted in the complete disappearance and apparent cure of an even larger number of cases of a similar type; but neither toxins nor radium, singly or combined, have effected a cure in any case associated with marked new bone production. Amputation followed by prolonged treatment with the mixed toxins in a series of thirty-eight consecutive cases shows 50 percent of the patients alive and well from three to eighteen years. This series includes all types of osteogenic sarcoma, as well as the cases associated with marked new bone formation. In a similar series of cases treated by amputation alone without toxins or radium before or after amputation, not a single patient has remained alive beyond the three-year period.

The choice of treatment in a given case depends on whether it belongs to the group of round cell sarcoma (endothelioma, according to Ewing's classification) or to the group associated with marked new bone formation. If to the first group, we believe it safe to try a combination of the systemic effect of toxins and the local effect of radium, the duration of the treatment to depend on the result obtained; if marked improvement is noted, the treatment may be continued until the tumor has entirely disappeared; but if no improvement is noted in from four to six weeks amputation should be performed, followed by prolonged systemic toxin treatment.

If the case belongs in the second group, we see no advantage, but a distinct disadvantage, in preliminary treatment with radium, roentgen rays or toxins, for the reason that metastases may develop during the period of treatment or may be hastened by the rapid breaking down of a vascular tumor, which permits living cells to be carried to the lungs or to remote parts of the body. As our experience, supported by the results of other men, shows that there is no reasonable hope of saving the limb in this group of cases, we believe that amputation at the earliest possible moment followed by prophylactic toxin treatment offers the greatest hope of saving the life of the patient.

The fact that ten inoperable cases of this series have been successfully treated with the mixed toxins alone or combined with radium, the patients remaining well from five to twenty-four years later, should prevent us from abandoning all hope in cases beyond surgical relief.

This article, a continuation of the article which appeared in the December 1926 issue of Archives of Surgery, contains summaries of fifty-four cases of bone sarcomas treated with Coley's mixed toxins or surgery and mixed toxins combined. Some of the cases were also treated with radium and/or amputation.

Selected Case Report

Case 1. Recurrent spindle cell sarcoma of tibia; amputation advised by other surgeons; treated with toxins alone; patient well, with a normal limb, at present, twenty-six and one half years later.

W. F., a man, aged 27, first noticed a swelling of the left tibia at the junction of the middle and upper thirds in March 1897; this slowly increased in size and, July 28, he was operated on by Dr. Stewart of Toronto, Canada. A prompt recurrence took place and, November 25 of the same year, a second operation was performed, consisting in incision and curetting of the bone: The tissue was sent to Dr. John Caven, professor of pathology, University of Toronto, who pronounced it a spindle cell sarcoma. The tumor again recurred, and the patient was referred to us for toxin treatment in February 1899.

Physical examination at this time showed a tumor at about the junction of the middle and upper thirds of the left tibia, measuring 3 by 4 inches (7.6 by 10.1 centimeters), with an ill-defined border; over the central portion were two ulcerations the size of a silver quarter. The patient was admitted to the Memorial Hospital where the mixed toxins of erysipelas and Bacillus prodigiosus (Coley) were begun at once and continued for about two months. The tumor rapidly disappeared and the bone cavity healed with healthy granulations. Before the healing process was entirely completed, the patient contracted a severe attack of accidental erysipelas (a patient with a fresh case of erysipelas had been in the ward not long before), which ran the usual course of about ten days. Healing continued rapidly and was completed two or three weeks later, after which the patient returned to his home in Canada. At the present time, twenty-six and one half years later, he is in excellent condition with a sound and useful leg, and is able to attend to his duties as a farmer.

In 1936, Dr. Coley reported that this patient has been followed up and is well thirty-seven years after original presentation.

Spontaneous Regression (Cure?) of a Malignant Tumor of Bone

Levin EJ

Cancer 10(2): March-April 1957; 377-381

Extracted Summary

A microscopically proved and radiologically confirmed malignant tumor of the humerus has healed spontaneously. Roentgenograms taken four years after the onset of symptoms demonstrate complete healing of the bone, and the patient is free of disease. There has been no therapeutic modality used, and an explanation of the cure has not been uncovered. The microscopic sections were submitted to many prominent pathologists, all of whom made an unequivocal diagnosis of malignant tumor, although there was considerable disagreement in naming the cellular type of the tumor.

Selected Case Report

The patient, a 29-year-old white woman, noted the gradual onset of pain in the left shoulder in June or early July of 1952. There was no known trauma or accompanying systemic abnormality, and her preceding state of health had been excellent. The pain was dull and aching in quality but never severe enough to warrant a medical consultation or even to justify medication.

On August 12, 1952, while hanging clothes, there was an acute exacerbation of the pain, followed by rapid swelling of the arm. The pain and most of the swelling subsided in a few days, leaving the arm bowed laterally. She was finally induced to see her physician, who made a tentative diagnosis of pathological fracture of the left humerus and referred her to St. Mary's Hospital, Cincinnati, for study and treatment.

On examination there was obvious swelling and deformity of the left arm. A large, firm, tender mass encircled the humerus, and there was questionable enlargement of the axillary nodes. No other subjective or objective abnormality was presented.

Since the birth of her fourth child in May 1951, there had been no pregnancies, menstrual abnormalities, or unusual vaginal discharge. The last child was born in St. Mary's Hospital, and although no note concerning the state of the placenta was found in the chart, her obstetrician recalled no unusual appearance of this structure.

Roentgenograms of the left arm on August 22, 1952, demonstrated extensive "moth-eaten" destructive changes involving the humerus from the surgical neck midway down the shaft. There was a pathological fracture with lateral angulation, but no evidence of callus formation, periosteal proliferation, or "tumor bone" was seen. A large soft tissue mass surrounded the area of involvement, and there was no soft tissue reticulation, ossification, or calcification.

A biopsy was obtained through a lateral approach, and a histological diagnosis of osteogenic sarcoma was considered most likely, although the possibility of malignant giant cell tumor could not be excluded. Although she was advised of the nature and prognosis of her disease, the patient refused all forms of definitive therapy. A hanging cast was applied and the patient discharged.

On September 29, 1952, roentgenograms of the arm demonstrated an increase in the amount of destruction. Periosteal proliferation was now seen, particularly on the lateral surface of the humerus adjacent to the fracture site. The soft tissue mass at this time was more distinctly outlined.

She returned home to take care of her children and experienced nothing more than slight pain in the arm despite her strenuous household duties. During the next few weeks even this slight pain subsided. The cast was removed about October 15, 1952. Nothing more was heard of the patient until she returned to her physician on January 25, 1955. She was in excellent health, without a complaint. The skin over her left humerus was the same color and texture as that over her right, showing only the biopsy scar. The arm was neither tender nor indurated, and no mass was palpable. She denied having undergone any form of therapy, either medicinal or physical. The only reason for the visit was the illness of her daughter.

A roentgenogram at this time showed complete healing of the humerus, except for residual slight deformity and sclerosis. A complete roentgenological survey in January 1955, and another in May 1956, including roentgenograms of the skull, chest, abdomen, and extremities, revealed no abnormality. No change in the humerus occurred in the interval between January 1955, and May 1956.

As of January 24, 1957, the patient is feeling fine, has no complaints, and is still unable to give any lead that would explain the apparent cure.

Spontaneous Regression of Reticulum-Cell Sarcoma of Bone

A Case Report

Cole RL; Ferguson MR

Journal of Bone and Joint Surgery (United States) 41-A(5): July 1959; 960-965

Extracted Summary

The purpose of this report is to present a case of primary reticulum-cell sarcoma of bone, proved by biopsy, which is unique in several respects. The patient, a five-year-old boy, is younger than any found reported in the literature. The youngest patient previously reported was an eight-year-old child in the series of Valls, Muscolo, and Schajowicz. This is also the only reported case of reticulum-cell sarcoma, with or without metastasis, which regressed spontaneously. Of reported cases, similar in location and histological appearance, which have regressed, particularly Case 5 of Magnus and Wood's series, Case 7 of Sherman and Snyder's report, and the case of sarcoma reported by Christian and Palmer, all had some form of therapy before regression.

Selected Case Report

A five-year-old Caucasian boy, R. J., was first seen at the Orthopaedic Hospital on November 1, 1951, complaining of pain and swelling of the outer aspect of the right leg. History revealed that the patient had struck his right leg approximately one month before while playing. He complained of pain only temporarily at that time and there was no noted swelling or discoloration. However, for two weeks prior to admission, the boy had complained of intermittent pain in the right leg; some six days preceding admission the mother had noted swelling about the painful area without increased heat but with slight pinkness on one occasion. It was further stated that the patient did not complain of pain on weight-bearing, but would not let anyone touch the leg because of pain. The past history revealed no serious illness, injuries, or operations.

Initial examination on admission revealed a well-developed, not acutely ill five-year-old boy with normal gait. Positive findings were limited to the right lower extremity where there was a firm swelling about the distal third of the fibula with some overlying skin warmth. There were no palpable lymph nodes in the groin or popliteal fossa on the right. The ankle joint adjacent to the swelling was not swollen or painful to movement.

Roentgenograms of the right leg made the same day showed a destructive lesion originating just proximal to the distal epiphysis of the fibula and extending upward for four and one-half inches with patchy destruction of cortex and medullary canal and a Codman's triangle along the medial cortex. A roentgenogram of the chest made at that time revealed the lung fields to be clear.

The white blood count was 12,800 with 54% polymorphonuclear neutrophils, 42% lymphocytes, and 4% eosinophils. The red blood count was 4,390,000 and the hemoglobin, 78%. The sedimentation rate (Cutler) was 18 mm/hour. Serum calcium (one determination) was 14 mg/100 cc, serum phosphorus was 4.1 mg/100 cc, and the alkaline phosphatase was 10.6 King-Armstrong units.

A working diagnosis was made at that time of a destructive lesion of the distal half of the right fibula consistent with Ewing's endothelioma, osteogenic sarcoma, or osteomyelitis.

On December 6, 1951, a biopsy was done which consisted of resection of the distal half of the diaphysis of the right fibula. At surgery, grayish tumor tissue was found which had infiltrated beyond the bone into the soft tissue about the lower end of the fibula. The cortex was almost completely destroyed, being replaced with necrotic gray material. The distal half of the fibula to the epiphyseal plate was resected with the attached soft tissue. However, because of the malignant appearance of the tumor and the working diagnosis, the procedure was confined to biopsy. No attempt was made to remove all evident tumor tissue, some of which was noted to remain in situ at the time of closing the wound. Surgical margin pathological studies were not attempted.

The biopsy report of Dr. Roy Hammack of Los Angeles, now deceased, noted that this was a very cellular tumor, the cells being round or oval with scanty, pale-staining cytoplasm and large, round or oval, vesiculating nuclei. Mitoses were found in moderate number. Supporting connective tissue strands contained blood vessels and occasionally the cells formed a radial arrangement about the strands. An occasional blood vessel wall appeared to be invaded by tumor. In some areas, muscle fibers were enclosed by tumor tissue. Areas of necrosis were frequent. The diagnosis was reticulum-cell sarcoma of the fibula and adjacent tissues.

The postoperative course was uneventful and the patient was discharged from the hospital on December 13, 1951, one week following surgery. On December 20, 1951, two weeks postoperatively, the sutures were removed

from the healed wound and it was recommended that the patient have roentgen therapy directed to the right distal fibular area. The parents decided to take the boy to the Mayo Clinic for evaluation. On January 9, 1952, a letter was received from the Mayo Clinic with the diagnosis of Grade IV, malignant, small cell neoplasm, probably atypical Ewing's tumor since the cells were larger than in typical Ewing's tumor. The working diagnosis was changed to Ewing's sarcoma and subsequent recommendations for therapy were based upon that diagnosis.

The patient returned to this hospital on January 24, 1952, asymptomatic except for minimal discomfort about the operative site. No positive clinical findings were noted. Roentgenograms of the right leg were made revealing the surgical resection of the distal shaft of the fibula which spared the epiphysis but included the original tumor area. The shaft of the fibula, proximal to the excised portion, showed evidence of internal erosion of cortex and mottled rarefactions in the medullary canal, findings consistent with tumor involvement. At that time, white blood count was 8,800 with 40% polymorphonuclear neutrophils, 50% lymphocytes, 4% transitional cells, 4% eosinophils and 2% basophils. Red blood count was 4,700,000. Hemoglobin was 80%.

A roentgenogram of the chest was made the following day which revealed multiple, small to medium sized rounded areas of infiltration scattered throughout the bases of both lung fields. Because of their bilateral homogeneous appearance, embolic lesions were considered most probable. Since there were no signs of infection, the lesions were thought to be other than septic in origin. Multiple infarcts might have been considered because the roundness of the infiltrates was atypical. The lack of pleural effusion would further indicate against such a diagnosis.

Because of the lack of pulmonary symptoms and the diagnosis of Ewing's sarcoma, the roentgenologist interpreted the findings as consistent with metastatic malignant disease. Palliative therapy was recommended since the diagnosis of Ewing's sarcoma was accepted and it was thought that local as well as metastatic pulmonary spread had already occurred. In order to prevent a large painful tumor during the terminal stages, resection of the remaining right fibula was recommended and roentgen therapy prior to surgery was suggested. The roentgenologist did not feel that roentgen therapy in the presence of pulmonary metastases would alter the clinical situation. Accordingly, roentgen therapy was not given. The local palliative surgery was not carried out and it was decided to continue conservative treatment only.

The patient was next seen on February 14, 1952, two months postoperatively, with a complaint of some local tenderness in the area of the scar. Objective findings were minimal and no therapy was instituted. On March 17, 1952, three months postoperatively, the boy was seen with an upper respiratory condition of approximately four days' duration. History revealed that it seemed to settle in the chest and cause a non-productive cough. A roentgenogram of the chest was made which revealed that the multiple infiltrates found in the previous roentgenogram of the chest had completely disappeared. The patient was then treated conservatively at that time and the cough disappeared in approximately two weeks.

At examination four months postoperatively there was no clinical evidence of local recurrence. Roentgenograms of the leg made then revealed that the distal end of the remaining upper fragment of the fibula had approached the normal.

During the remainder of 1952 the patient's general health remained excellent and there was no clinical evidence of tumor recurrence. Periodic roentgenograms of the right leg made during that year revealed, by ten months postoperatively, that the bone structure of the tibia and remaining fibula was normal. Similarly, frequent roentgenograms of the chest failed to reveal any recurrence of the pulmonary findings. At that time, the case was reviewed and the working diagnosis was changed to reticulum-cell sarcoma with regression of local and metastatic disease.

The patient has been followed at yearly intervals with physical examinations and roentgenograms of the right leg and chest. He was last seen in February 1958, six years postoperatively, at which time a complete skeletal survey was performed in addition to physical examination. His health has remained good and he has remained free of recurrence of the original reticulum-cell sarcoma of the distal portion of the right fibula, clinically and roentgenographically.

Photomicrographs of the biopsy material demonstrate the characteristic cellularity, alveolar arrangement of the cells, and the round or oval reticulum cells with scanty, pale-staining cytoplasm and large, round or oval, indented or lobulated, nuclei with prominent nucleoli and coarse chromatin.

In 1957, more than five years after the biopsy was reported and because of the benign course, the original slide and new slides made from the biopsy were reviewed by Dr. J. Vernon Luck and Dr. Weldon K. Bullock of the Orthopaedic Hospital, Los Angeles, and by Dr. Fred W. Stewart of the New York Memorial Hospital. All confirmed the diagnosis of reticulum-cell sarcoma of bone.

Spontaneous Regression of a Malignant Primary Bone Tumour

Dissing I; Heerfordt J; Schiodt T; Sneppen O
Acta Orthopaedica Scandinavica 49(1): 1978; 49-53

Extracted Summary

A histologically confirmed malignant, primary bone tumour in the pelvis, presumably an osteosarcoma, underwent spontaneous regression. The large tumor was inoperable and gave rise to severe pain as well as difficulty in walking. After 2 years of progression, with increasing destruction of the pelvic bones, the clinical and radiological condition improved spontaneously, and at present the patient is alive, almost symptom-free, after 6 years of follow-up.

Selected Case Report

The patient, a 36-year-old man, was first seen in February 1971. Since 1968 he had been suffering from increasing pain on weight-bearing in the region of the left hip, occasionally radiating to the leg. His general condition was good, and, except for mild atrophy of the muscles of the left thigh and a limp, physical examination showed no abnormalities. Radiography disclosed a widespread osteolytic lesion in the left half of the pelvis. Chest radiography was normal. ESR was 100 millimeters and alkaline phosphatase was slightly elevated. Open biopsy was done at the anterior demarcation of the tumour in the iliac bone. It showed, as did subsequent revisions of the same preparations, benign chondroblastoma. In view of the clinical and radiological findings, however, this diagnosis did not seem convincing. Therefore, an exploratory operation through a wide posterior approach was performed in March 1971. In the iliac bone there was a tumour about 12 x 8 x 8 centimeters, anteriorly involving the superior ramus of the ischium and posteriorly the sacroiliac joint and the lateral mass of the sacral bone. Ample tumour tissue was excised for histological examination which showed, as did subsequent histological revisions of these preparations, a malignant, primary bone tumour, presumably a highly differentiated chondroblastic osteosarcoma, although the possibility of malignant transformation of a chondroblastoma could not be ruled out. It was now evident that the tumour was inoperable, and, as there did not seem to be any indication for radiotherapy or chemotherapy, the patient received only symptomatic treatment, viz., non-weight-bearing and analgesic medication.

Over the next few years the patient, still untreated, was followed up in the outpatient clinic. Until the spring of 1973 his condition deteriorated steadily. The regional pain grew worse, and now he also had rest pain. The left leg became 2 centimeters shorter and the difficulty in walking increased, necessitating the use of two English canes. Radiographic examinations during this period showed increasing destruction of the left half of the pelvis. But in the course of the latter half of 1973 a reversal occurred. The pain decreased, and during the following years the complaints gradually disappeared. Since 1975 the patient has been free of pain, and he walks almost normally. Accompanying the clinical improvement a radiological remission took place. The tumour area became increasingly delimited and sclerosed. When last seen, in March 1977, the patient was feeling perfectly well. There have not at any time been signs of dissemination of the tumour, in particular not to the lungs.

Regression of Osteogenic Sarcoma Metastases Associated with Intensive Meditation

Meares A
Medical Journal of Australia 2: Oct 21 1978; 433

Extracted Summary

The patient described showed marked regression of metastases associated with intensive meditation. It would seem that the patient has let the effects of the intense and prolonged meditation enter into his whole experience of life. His extraordinarily low level of anxiety is obvious to the most casual observer. It is suggested that this has enhanced the activity of his immune system by reducing his level of cortisone.

Selected Case Report

The patient, aged 25, underwent a mid-thigh amputation for osteogenic sarcoma 11 months before he first saw me 2 1/2 years ago. He had visible bony lumps of about 2 centimeters in diameter growing from the ribs, sternum and the crest of the ilium, and was coughing up small quantities of blood in which, he said, he could feel small spicules of bone. There were gross opacities in the X-ray films of his lungs. The patient had been told by a specialist that he had only two or three weeks to live, but in virtue of his profession he was already well aware of the pathology and prognosis of his condition. Now, 2 1/2 years later, he has moved to another State to resume his former occupation.

This young man has an extraordinary will to live, and has sought help from all the alternatives to orthodox medicine which were available to him. These have included acupuncture, massage, several sessions with Philippine faith healers, laying on of hands and yoga in an Indian ashram. He had short sessions of radiation therapy, and chemotherapy, but declined to continue treatment. He has also persisted with the dietary and enema treatment described by Max Gerson, the German physician, who gained some notoriety for this type of treatment in America in the 1940s. However, in addition to all these measures to gain relief, the patient has consistently maintained a rigorous discipline of intensive meditation as described previously. He has, in fact, consistently meditated from one to three hours daily.

Two other factors seem to be important. He has had extraordinary help and support from his girl friend, who more recently became his wife. She is extremely sensitive to his feelings and needs, and has spent hours in aiding his meditation and healing with massage and laying on of hands.

The other important factor would seem to be the patient's own state of mind. He has developed a degree of calm about him which I have rarely observed in anyone, even in oriental mystics with whom I have had some considerable experience. When asked to what he attributes the regression of metastases, he answers in some such terms as: "I really think it is our life, the way we experience our life."

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Journal of Oral and Maxillofacial Surgery 45(1): Jan 1987; 53-57

Extracted Summary

This paper presents a case of benign osteoblastoma that showed regression during a long-term follow-up, and illustrates that the course of any disease in any instance is not always predictable with certainty. There are two aspects of this case that merit discussion: 1) the decision to defer intervention despite occasional reports of malignant transformation of osteoblastoma, and 2) the apparent spontaneous regression which followed biopsy, a phenomenon which to our knowledge has not been reported previously for osteoblastoma.

Selected Case Report

The patient, an 11-year-old Caucasian female, was first seen in 1971 because of a slowly increasing swelling of the labial aspect of the anterior mandible. Clinical examination disclosed prominence in the region of the left lateral incisor and canine. The enlargement was circular and measured approximately 7 millimeters in diameter. The overlying gingiva was normal in color and texture but the area was markedly tender to palpation. The four lower incisors were slightly mobile. All teeth in the area tested vital. The patient's medical history was negative and the remainder of the physical was within normal limits.

Periapical radiographs showed diffuse mottling of the alveolar bone in the lower anterior region extending between and below the roots of the incisor teeth. The periodontal ligaments in the involved area were thickened. Laterally and inferiorly the abnormal bone pattern blended imperceptibly with normal bone. Occlusal radiographs showed a small expansion of the mandible labially in the left canine region.

An incisional biopsy was performed under local anesthesia. No effort was made to remove the entire lesion. The specimen measured 5 millimeters in its widest diameter and included a portion of the labial cortex as well as a small sample of the underlying osseous tissue. Microscopic examination showed an irregular pattern of active bone deposition and resorption with numerous reversal lines. The marrow was highly vascular and richly cellular, with plump osteoblasts and numerous osteoclasts. Local pathologists and several outside consultants concurred in the diagnosis of benign osteoblastoma.

In the absence of a defined border in the radiographs it was felt that a distinct plane along which the lesion would separate might not be found on operation. In such circumstances surgical management would have required extraction of six lower anterior teeth and en bloc resection. The prospect of significant deformity led to several consultations culminating in the decision to defer surgery, and to monitor the status of the lesion for an indefinite period, leaving open the option to operate.

In subsequent months it became apparent that the process was not progressing. Serial radiographs taken at yearly intervals revealed that the process was a regressing one, marked by alterations in trabecular pattern and gradual restitution of normal architecture. Over a period of 15 years the lesion regressed to the point where the configuration was virtually normal, with reconstitution of the lamina dura and normal periodontal ligaments. External swelling has subsided, the teeth are firm and the patient is asymptomatic. If there are microscopic foci of disease still present, they are not detectable on clinical and radiographic examination.

Benign Osteoblastoma of the Spine

Report of Four Cases with One Case of Spontaneous Cure

Collignon JC; Kalangu K; Flandroy P

Neuro-Chirurgie 34(4): 1988; 262-70

Extracted Summary

In a six-year period, the authors have seen 4 cases of benign osteoblastoma of the spine. Two were located in the cervical spine, one in the thoracic spine and the fourth in the lumbar segment. Three were operated and cured after a follow-up from two to three and one-half years. One case spontaneously cured after a follow-up of five years. The roentgenographic features of these lesions vary according to their age, localization, biological behaviour and the pattern of their osseous host. Computed tomography is the best method to specify the site of the nidal lesion the total removal of which led to a complete cure.

Selected Case Report

The authors' first case concerns a 10-year-old girl who can be considered as the first example ever presented of spontaneous remission of an osteoblastoma. By making reference to the dimensional criteria of a nidal lesion, as mentioned above, we notice that this has a diameter of 1 centimeter in the osteoid osteoma; tomography of an osteoblastoma, instead, demonstrated a lesion 2 centimeters in diameter and involving the pedicle, pars interarticularis, inferior articular facet, and the tubercle of the tenth right rib. The lesion contained calcified elements and was surrounded by sclerosis of the vertebral body and of the lamina.

A clinical examination five years later demonstrated complete remission of the symptoms, and a CT scan demonstrated the right intervertebral foramen of Th10 and Th11 to be surrounded by sclerotic bone tissue in which no nidal lesions were visible. The reason for this favorable evolution is unknown.

(Noetic Sciences translation)

Osteochondromas

Spontaneous Resolution of an Osteochondroma

Callan JE; Wood VE

Journal of Bone and Joint Surgery (United States) 57-A(5): July 1975; 723

Extracted Summary

There have been numerous reports of osteochondroma in varied and sometimes unlikely locations, but to our knowledge, after a careful search of the English literature, spontaneous disappearances of osteochondroma has never been reported. We report a case of spontaneous disappearance of an osteochondroma in a five-year-old girl.

Selected Case Report

A five-year-old white girl, T. G., was first seen in our clinic in October 1972. While playing with her, her mother had noticed a hard lump in the proximal part of the left humerus. No associated clinical symptoms were present. The patient had had no operations, significant illnesses, or injuries. Physical examination was negative except for a solitary lump located on the posteromedial aspect of the humerus at the junction of the proximal and middle thirds. The lump was hard, nontender, and non-mobile. Roentgenograms of the left humerus showed a typical osteochondroma. Subsequent roentgenograms and clinical examination on July 10, 1973, revealed little change. On re-examination of the patient on November 6, 1974, however, the lump was no longer palpable. Roentgenograms showed the lesion to have nearly disappeared.

The "Disappearing" Osteochondroma

Paling MR

Skeletal Radiology 10: 1983; 40-42

Extracted Summary

Spontaneous regression of a solitary osteochondroma is described. This regression apparently resulted from a cessation of growth of the osteochondroma prior to skeletal maturation, with subsequent incorporation of the lesion into the enlarging bony metaphysis.

Selected Case Report

A 9-year-old boy presented with a palpable mass above the left knee. Radiographs demonstrated a solitary osteochondroma arising from the medial aspect of the distal metaphysis of the femur. The lesion was followed with annual radiographs, over a six-year period, without treatment. The ossified portion of the osteochondroma was observed to grow rapidly during the first two years. A short interval of apparent stability was then followed by progressive reduction in size of the lesion.

Spontaneous Regression of Osteochondromas

Two Case Reports

Copeland RL; Meehan PL; Morrissy RT

Journal of Bone and Joint Surgery (United States) 67-A(6): July 1985; 971-973

Extracted Summary

Much has been written on the subject of osteochondromas, but in the English literature there has been only one report of an osteochondroma that regressed spontaneously. For that reason, we report two additional cases of this phenomenon.

Selected Case Report

Case 1: This boy was eleven years old when radiographs were made of the knee after an injury. A solitary sessile osteochondroma was noted on the posteromedial aspect of the distal femoral metaphysis. It measured six centimeters in diameter at the base and projected 1.7 centimeters away from the cortex. The patient had been asymptomatic with regard to the osteochondroma, although the osseous projection was palpable. No therapy was recommended. Four months later, the mass was found to be reduced in size both clinically and on radiographs. Four months after that no mass in the thigh could be palpated and the radiographs revealed only a slight cortical prominence at the previous site of the lesion. At the last evaluation, nineteen months after the first presentation, the lesion could not be identified on the radiograph.

Resolving Solitary Osteochondromas

A Report of Two Cases and Literature Review

Montgomery DM; LaMont RL

Orthopedics 12(6): Jun 1989; 861-863

Extracted Summary

Resolving osteochondromas were previously thought to be uncommon. Two cases are presented, including the first report of a resolving solitary osteochondroma of the proximal tibia. These cases, along with previous reports, suggest resolution may occur, especially in boys under age 12 years. Surgical excision should be delayed in this group of patients provided malignant transformation or neurovascular compromise is not a concern.

Selected Case Report

In Case 1, an 11-year-old boy presented with a one month history of a nonpainful mass in the proximal portion of his left arm. He had no other palpable masses and no family history of osteochondromas. Radiographs showed a sessile osteochondroma of the humerus. Twenty-one months later, the mass was still palpable but appeared smaller on the radiograph. At 32 months, the mass was difficult to palpate. Radiographs showed continued resolution of the tumor. By 68 months, the mass was no longer palpable. Only a small osteochondroma remnant and slight widening of the shaft remained.

Malignant Neoplasms of Connective and Other Soft Tissue

Inoperable Sarcoma Cured by Mixed Toxines of Erysipelas

Wyeth JA; McCosh AJ

Annals of Surgery 25: 1897; 174-178

Extracted Summary

The topic of this surgical clinic is the use of mixed toxins of erysipelas in the treatment of sarcomas. Dr. William Coley presented a case of inoperable sarcoma of the shoulder in which mixed toxins of erysipelas produced a "cure."

Selected Case Report

Dr. W. B. Coley presented for examination through the permission of Dr. M. Storrs, surgeon to the Hartford Hospital, and his associate, Dr. R. S. Griswold, by whom she had been treated personally, a woman, forty-two years old; married. Her paternal grandmother died of cancer of the breast. She herself had always been well until the fall of 1895, when she first noticed a hard lump a little below the left clavicle. This grew rapidly in size, and early in December 1895, she consulted Dr. Storrs, of Hartford. The tumor then had reached the size of a small orange, and extended into the axilla, and was so adherent to the vessels that after a consultation with Dr. McKnight, one of the other attending surgeons to the Hartford Hospital, it was decided to be entirely inoperable. The patient was rapidly losing flesh and strength. As the case was regarded as hopeless, it was decided to try the erysipelas toxines for a short time. A preparation of the mixed toxines of erysipelas and bacillus prodigiosus, made at the Loomis Laboratory by Mr. B. H. Buxton, was obtained, and the injections were begun December 16, 1895, as directed by Dr. Coley.

Small doses were given, and no chill was obtained until December 29. The injections were given daily or at intervals of two days up to February 8, forty-eight injections having been given with a result of fourteen distinct chills; the dose was eight minims of the mixed unfiltered toxines.

On February 9 a stronger solution from more virulent cultures was obtained, which produced severe reactions in doses of two or three minims. The treatment was discontinued on March 17, 1896, since which time no further treatment has been given.

Soon after the treatment was begun the tumor began to shrink in size, and later to break down in places. This process became more rapid after the stronger solution was begun. By the latter part of March, 1896, the tumor had entirely disappeared, the patient had begun to gain rapidly in flesh and strength, and in a few months had entirely regained her twenty-five pounds of lost weight. She is at present in perfect health, and careful examination fails to reveal any trace of the tumor or any glandular enlargement.

A Case of Recurrent Sarcoma with Apparently Spontaneous Cure and Gradual Shrinking of the Tumour

Watson AL

Lancet 1: Feb 1 1902; 300-301

Extracted Summary

A case is presented of a spontaneous cure of a recurrent sarcoma of the back. No injections of mixed toxins were used nor did a spontaneous attack of erysipelas occur, yet, the tumor shrank in a most remarkable manner, and so far as its malignancy is concerned may be regarded as cured.

Selected Case Report

The patient, a strong, healthy-looking woman, aged 39 years, married, and with two children, was admitted to the Glasgow Royal Infirmary on November 21st, 1900. She was absolutely free from ailment, and came into the hospital simply because a swelling on the back was annoying her and she wished to have it removed. This tumour was large and pendulous, being attached to the left side of the back to a base nine inches wide and rather less than four inches in vertical measurement; the apex hung down for almost four inches below the lower margin of its attachment to the back while the upper margin was just below the inferior scapular angle. It was soft, but not fluctuant, slightly nodulated, and had the appearance of a lipoma undergoing degeneration. The skin covering the dependent portion was of a dark reddish colour. The tumour was regarded as simple, partly because of its character and partly because its history went back to the patient's childhood, though it had grown considerably in recent years.

On November 23rd Dr. D. McK. Dewar, acting for Dr. Newman, removed it freely with all the discoloured skin, and remarked at the time that in three respects it differed from an ordinary lipoma: (1) the tissue was of a dirty greyish yellow colour and very friable; (2) it was exceedingly vascular; and (3) its margins were diffuse and indefinite. After the operation hemorrhage was so free that the dressings had to be frequently changed in the first few hours; thereafter they were left for four days untouched. On the 27th it was observed that the dressings had become tight and on exposing the back the tumour was found to be almost as large as before operation. In the expectation that this might be due to accumulation of blood clot beneath the flaps a portion of the wound was opened up, but no blood could be expressed, even when a director was introduced and worked about inside the wound. It was therefore concluded that the swelling was due to a rapid reformation of the tumour. By an unfortunate oversight the tumour first removed had been destroyed without

microscopical examination; but at this stage two small portions of the tissue were removed. One of these was hardened and stained myself, the other was sent to Dr. C. Workman, pathologist to the infirmary. The sections obtained from the first portion were examined by Dr. Newman, by Dr. Dewar, and by myself; and the recognition of sarcomatous tissue, chiefly round-celled, was unhesitating. Dr. Workman's quite independent report described the tissue as a mixed round cell and spindle cell sarcoma.

A week later the operation wound had begun to ulcerate along its margins, and it was decided to attempt a wide removal, but when on December 4th the patient was placed on the table and anaesthetized, the tumour had attained such a size and was so diffuse in its margins that further radical interference was considered out of the question; the ulcerated edges were therefore cut away and the wound was again stitched up. During the remaining days of her residence in the infirmary the patient looked and felt very ill and weak. A bad prognosis was given and she was dismissed on the 13th. After this several medical men saw her and all took a gloomy view of the case; but in spite of everything she improved and a month after dismissal from the hospital a letter was received from her own medical attendant stating that the operation wound had almost completely healed. When I visited her at her home in the country on February 9th, 1901, I reported as follows: "The operation wound has entirely healed up, with only a small scab at his lower extremity, and leaves a firm healthy looking cicatrix. The tumour is much less tense than at the time of dismissal and shows a tendency to return to the pendulous form which it had when first seen; it is flaccid and quite painless. At its anterior margin are some subcutaneous livid patches which were noticed at the time of the recurrence of the tumour, but these do not appear to have changed at all or to be spreading. The patient's general condition is much improved; she is able to be up and about, eats with good appetite, and in general feels very comfortable." She again reported herself at the infirmary in April and the tumour seemed to be still further diminished in size.

As I have already stated, there was nothing of the nature of an attack of erysipelas in this case; but although locally no inflammatory phenomena manifested themselves, the details of the temperature chart are suggestive. For the first six days after admission the temperature records showed a subnormal and normal range with a minimum of 96.8° F. and a maximum of 98.6°F., being quite unaffected by the operation on November 23rd, 1900. Then during the following week a slight upward tendency was noted, a maximum of 99.8° being registered on December 3rd, and after the second time to the theatre (on the 4th) the temperature shot up to 101°, remaining febrile and subfebrile, with daily remission for five days, the highest point reached being 102° on the 8th. From the 10th to dismissal on the 13th it was practically normal. Whether this postoperative fever was an important factor in deterring the subsequent resolution of the tumour I cannot pretend to say, but certainly the progress was continuous and most satisfactory.

Finally, I again visited the patient at her home on November 21st, exactly a year after her admission to the infirmary, and the following is my report: "The general health is excellent. The tumour is much less prominent and hangs in a loose sac, as if almost all its contents had been evacuated. There is a slight lividity at the margins but no pain, except an occasional stinging like neuralgia in damp weather. The tumour covers about the same area as at the time of dismissal but consists of little more than loose skin. The scar of the operation wound is perfectly sound and firm. No pain is caused by free manipulation and the clothes are worn without discomfort."

Spontaneous Cure of Congenital Recurring Connective Tissue Tumor

Shore BR

American Journal of Cancer 27: 1936; 736-739

Extracted Summary

A spontaneous cure of a congenital, recurring, supposedly malignant connective tissue tumor of the interscapular region of a six-month-old child is reported. The diagnosis was based upon the histologic study of two biopsy specimens in two different laboratories. The growth disappeared following the second imcomplete operation, and the child has remained well for seven years.

Selected Case Report

The patient, a six-month-old girl, was admitted to St. Luke's Hosptial on November 20, 1929, with the diagnosis of recurrent tumor of the interscapular region. The mother stated that the child was born with a tumor on the back. The growth increased in size and was removed surgically by the obstetrician when the baby was two and one-half months old. The tissue was sent to the State Institute for the Study of Malignant Disease in Buffalo, New York, where the diagnosis of fibrosarcoma was made. Within two weeks following the excision of

the growth, a recurrence was discovered beneath the scar: this persisted and increased in size.

Examination showed a normally developed girl without congenital abnormalities. In the midline of the back at the level of the scapulae was a raised mass about 5 centimeters in diameter, fixed to the overlying skin by a transverse, healed surgical scar. The tumor was firm, movable with the skin and underlying muscles, and was not tender.

The diagnosis of recurrent, operable fibrosarcoma was made and surgical removal attempted. At the time of operation, however, tumor tissue was found widely infiltrating the underlying muscles and overlying skin, and complete excision was impossible. A wedge-shaped piece was removed from the center of the growth for histologic study, the bulk of the tumor being left in situ. The wound edges were united with interrupted horsehair sutures.

Pathologic Report (by Doctor L. C. Knox): The specimen consists of a piece of skin and tumor from the interscapular region. The skin is triangular in shape and measures 4 x 4.5 x 2.2 centimeters. It overlies a mass of opaque, homogeneous, white tumor tissue which is 2 centimeters thick. Histologic study shows a portion of the skin surface with relatively normal epithelium and corium. The superficial layers of the fibrous tissue are also normal. Beneath this is a solid mass of tumor invading the fat and muscle. It is of the connective tissue type, is not extremely cellular, and is undergoing both hyaline and myxomatous degeneration. The nuclei are elongated, rather plump, and are not extremely hyperchromatic; mitoses are not numerous. The tumor is of the type, however, which is likely to recur even though thoroughly removed. Diagnosis: fibrosarcoma, possibly of a neurogenic origin.

Subsequent Course: The wound healed by first intention and the patient was discharged from the hospital on the eleventh postoperative day. A letter from the mother, dated March 23, 1930, stated that the child was well and that the growth was slightly larger than it was when she had left the hospital three months previously. At her first follow-up visit, on July 29, 1930, the mass had decreased in size. The improvement was spontaneous and had occurred in the absence of any further surgical, radiation, or medicinal therapy. The patient was pale but was growing normally and appeared to be in good health. A letter dated January 27, 1931, thirteen months after operation, stated that the condition was about the same as in July. On May 29, 1931, the patient was examined and appeared healthy except for slight pallor. There was no evidence of residual or recurrent tumor. At that time the mother stated that the tumor had disappeared and reappeared beneath the scar several times during the past year. However, no return of the growth has been noted since the patient was seen on May 29, 1931. Roentgenograms of the chest made on January 26, 1932, showed normal lung fields with no evidence of metastatic tumor.

Letters from the mother dated April 26, 1933, and November 23, 1933, stated that the child was well with no evidence of tumor. At her last examination, August 23, 1934, almost five years after operation, there was no sign of residual or recurrent growth. A letter of June 23, 1936, seven years after operation, stated that the child was well and had no complications except occasional itching in the region of the scar.

Spontaneous Regression of a Case of Myosarcoma

Penner DW

Cancer 6(4): July 1953; 776-779

Extracted Summary

A case of spontaneous regression of a myosarcoma of the lower end of the thigh in a 2-month-old white male child is presented. The patient is alive, with no evidence of tumour, five and a half years after biopsy. No treatment of any type was used. It is doubted if the biopsy, which was small and superficial, would have produced sufficient interference with the blood supply to have caused regression of the tumour. From the mother's observation, no change in the size of the tumour was noted until three months after the biopsy, and then, in the next four months, the tumour gradually decreased in size and disappeared. Five years later, no evidence of tumour remains, and the absorption of bone seen at the lower end of the femur has disappeared, the bone now being normal.

Selected Case Report

On June 22, 1947, a 2 1/2-month-old white male baby, H. C., was brought by his mother to a physician two weeks after she noted a tumour located on the lateral surface of the left thigh, just above the knee joint. The family physician confirmed the presence of a painless swelling above the left knee and also discovered a swelling of the left sternomastoid muscle. The latter was diagnosed as a torticollis. The patient was then referred to Winnipeg and admitted to the General Hospital on June 22, 1947. A tumour, 5 centimeters above the knee joint,

located on the anterolateral surface of the left thigh, was present. This was firm and measured 5 centimeters in diameter. The mass was fixed to surrounding tissue; the overlying skin was intact.

Roentgenograms of the left thigh: "the left femur shows a crescentic defect involving the lateral condyle and part of the shaft of the distal end of the femur. There appears to be a soft tissue tumour in the lower aspect of the thigh. Summary: soft-tissue tumour eroding the lower end of the femur."

The baby's weight in the hospital was 8 pounds, 12 1/2 ounces. No laboratory procedure other than a routine urinalysis was done. This was normal. A left sternomastoid swelling was noted. On June 24, 1947, under a general anesthetic, a biopsy of the tumour was performed. The exposed tumour lay deep to the fascia. The cut surface of the tumour, as seen during removal of biopsy, was homogeneous and light pink in colour and, in the gross, was considered to be sarcoma by both the attending surgeon and the writer.

Pathological Report on the Biopsy. Gross Findings: Two pieces of the homogeneous light pink coloured tissue, measuring 1.2 centimeters in diameter. Microscopic Findings: This was a fairly vascular, cellular tumour. Most of the cells were uniform in size and staining reaction. The nuclei were elongated with rounded ends. The cytoplasm was scanty, with indistinct cell margins. Mitoses were fairly frequent, with up to four per high power field. Classification of this tumour was not definite, but because of a small amount of glycogen in the cytoplasm a diagnosis of myosarcoma was suggested. A decision was made against further treatment. The baby was discharged from the hospital on July 2, 1947. The parents were told that the infant had a malignant tumor. The diagnosis of sarcoma was confirmed by Fred W. Stewart and Frank W. Foote, Jr., Memorial Hospital, New York City.

Late in 1951, the Manitoba Cancer Institute Follow-up Service, after having "lost" the case for a number of years, finally managed to contact the mother and found that the child was alive and well. After considerable delay, the child was brought to Winnipeg on September 5, 1952. Complete investigation showed a well-nourished and developed, bright, 5 1/2-year-old white boy with no evidence of tumour. All that remained was a linear 12.5 centimeter scar on the anterolateral aspect of the thigh, extending upwards from the superior border of the patella. Detailed questioning of the mother, who is intelligent and cooperative, revealed that the tumour of the thigh remained the same size until the age of 5 months, when it began to decrease in size. The swelling was completely gone by the time the child was 9 months of age.

Every aspect of the child's environment, family history, diet, and development was reviewed. Nothing of note was found. The child had seen no other doctor and had not been taken to any irregular practitioner. No form of local or internal medicine was ever used. The child's diet was well-balanced with no irregularity. The father, who is a hard-rock miner in Northern Manitoba, has an average middle-class income. Only twice, for brief visits, was the child away from his home. This boy began to walk at 13 months and was never seen to favor either leg, nor has he ever complained of local pain.

On physical examination in September 1952, his height was 44 1/2 inches, weight was 45 pounds. Some atrophy of the upper end of the left sternomastoid muscle was present. The extremities showed a scar, 12.5 centimeters in length, on the anterolateral surface of the left thigh, extending from the superior border of the patella. No mass was palpable deep to the scar. The circumference at the upper border of the patella was 0.8 centimeters greater on the left side. Normal and full range of movement were present at all joints. There was no pain or tenderness. Gait was normal.

Laboratory Investigation, September 1952. A roentgenogram of the chest was normal. Roentgenograms of the pelvis and long bones of both lower limbs showed no abnormality of the bone or soft tissue. The urinalysis was normal. The hemoglobin was 12.6 gm/100 ml, and the red blood count was 4.35 million/mm^3. The white blood count was 6,100, with a normal differential. The sedimentation rate was 16 millimeters in a 200 millimeter tube per hour. The biochemical analysis showed the serum calcium to be 10.4 mg/100ml; the serum alkaline phosphatase, 26.7 King units; the serum acid phosphatase, 5.1 King units; the serum phosphate, 4.8 mg/100 ml; and there was a total of 7.7 grams proteins, of which 5.2 grams was albumin and 2.5 grams globulin.

Spontaneous Regression of Malignant Tumors

Report of a Twelve-Year Spontaneous Complete Regression of an Extensive Fibrosarcoma, With Speculations about Regression and Dormancy

Dobson L; Dickey LB

American Journal of Surgery 92: Aug 1956; 162-173

Extracted Summary

Malignant tumors frequently show fluctuation in rate of growth. Some have periods of very slow and then rapid growth. Some are slow to metastasize or the metastases may be dormant for many years.

There are a few proved cases of malignant tumors that have completely regressed without any therapy. Most of the cases reported as spontaneous cures cannot be accepted when examined critically. To aid in the segregation of cases of complete spontaneous regression, a list of criteria is proposed.

A case is reported of a girl who had an extensive fibrosarcoma when five months of age, received no treatment and is now entirely well at thirteen years of age. A list of possible factors concerned in spontaneous regression of malignant tumors is presented. The various theories which have been suggested to explain late recurrence and metastasis are discussed. The variability of growth of all types of malignant tumors must be considered when evaluating a particular type of therapy or in estimating the prognosis.

Selected Case Report

Patient S.V. was first seen in Stanford Hospital on May 5, 1943, at the age of five months, because of a large tumor in the left upper thigh and lower abdomen. When the baby was two months old, her mother had noticed a lump in the left inguinal fold. This mass had grown steadily, had bulged out the inguinal fold and had produced a marked swelling of the upper left thigh. She had been a feeding problem since birth, with a great deal of colic and constipation. Several days before admission she began to have frequent green, watery stools containing undigested food.

There was no fever on admission but on the fifth and sixth days her temperature rose to 38.5°C. Examination showed a well-nourished, well-developed baby five months of age. The upper left thigh was about 50% larger than the right, and on palpation there was a 9 by 7 centimeter rather soft, deep-seated mass extending downward below the left inguinal ligament. Abdominal palpation revealed a firm, fixed mass extending upward from the left iliac fossa laterally in the flank to above the left costal margin. The mass in the thigh seemed to be an extension of the abdominal mass.

Laboratory studies included the following: Blood counts: May 7, 1943; erythrocytes 4,320,000, hemoglobin 14 gm%, 69% Sahli, leukocytes 7,400, neutrophils 41%, eosinophils 2%, lymphocytes 51%, monocytes 6%. May 11, 1943; leukocytes 11,600, neutrophils 46%, eosinophils 1%, lymphocytes 53%. Urinalysis: May 7, 1943; no albumin, no sugar, no casts, rare red blood cells, 2 to 4 white blood cells per high power field. Tuberculin test: May 10, 1943; 1/1000 OT. negative at twenty-four and forty-eight hours. Sedimentation rate: May 10, 1943-22 millimeters in 60 minutes (Wintrobe). Stool culture: May 10, 1943; "No members of the typhoid, paratyphoid, or dysentery group were isolated." X-ray examinations: May 7, 1943, the patient's chest was clear. The soft tissue mass was noted in left upper thigh. Excretory urograms showed both kidneys were functioning well. There was a large mass filling the left flank and iliac fossa, displacing the descending colon and small bowel to the right, and pressing on the left side of the bladder. The diagnoses considered at that time were: (1) sarcoma of some type, (2) hemangioma or lymphangioma and (3) lipoma.

A few days later the inguinal mass was aspirated to rule out a psoas tuberculous abscess but no pus was found. On May 15th the mass in the thigh was biopsied through a long incision. It was planned that if the tumor should prove to be benign, the incision would be extended upward across the inguinal ligament and flank. A diffuse, pink, vascular tumor was encountered which was invading muscle and had surrounded femoral vessels and nerve. A frozen section was examined and found to be a malignant tumor, some type of a spindle cell sarcoma. Since it was impossible to remove the tumor, the wound was closed.

The report on the permanent sections and special stains is as follows: "Gross Specimen: Consists of a small wedge-shaped piece of pale grayish-white tissue which has the appearance of fish flesh. It measures 1 x 0.8 x 0.4 centimeters, and is soft and semitranslucent."

"Histological Examination: Sections show tumor of rather uniform structure consisting of loosely grouped, small spindle shaped tumor cells with small, oval to polygonal nuclei. There is considerable fine fibrillary material and reticulated stroma between the tumor cells. Mitotic figures are not numerous. Van Gieson's stain shows slender fibers of collagen between the tumor cells. Mucicarmine stains reveal nothing in the matrix indicative of mucin. The diagnosis is sarcoma, thigh, fascial (fibrosarcoma)."

The subsequent week the case was presented to the Stanford Tumor Board. It was decided not to use x-ray therapy because the sarcoma was well differentiated and unlikely to respond to radiation; a high dose over so large an area in the abdomen and thigh could not be borne. A safe dose would have been ineffective and would merely have made the baby very sick.

The baby's temperature rose to 38.8°C. for two days postoperatively, then dropped to normal. No treatment was given unless the operation for the biopsy under general anesthesia and several x-ray examinations can be considered forms of therapy.

The skin sutures were removed on the seventh postoperative day and the baby was sent home with the wound healing satisfactorily. Her parents were given a hopeless prognosis. We expected that within a short time the rapidly growing sarcoma would produce a fungating mass in the groin or flank.

Two months after operation, however, we found the wound well healed and the tumor no larger. Four months postoperatively we thought the abdominal and thigh masses were both a little smaller.

The child was followed up regularly and the masses continued to recede until eighteen months after the biopsy when no masses were palpable. She had begun to walk at thirteen months of age. We have seen her at yearly intervals. X-ray examinations have been made many times, the last being in July 1955. On five occasions she has been presented before the Stanford Tumor Board as having a rare case of spontaneous regression of a malignant tumor.

The microscopic sections have been examined by at least fifteen pathologists in two medical schools. New sections have been cut and special stains made on several occasions. All pathologists agree on the diagnosis: malignant fibrosarcoma.

In studying the case report of Cushing and Wohlbach (American Journal of Pathology 3 (1927) 203-217) it was noted that the biopsy specimen of the malignant paravertebral sympathicoblastoma had been diagnosed as a spindle celled sarcoma. That child was treated with Coley's toxin. Ten years later a laminectomy was performed and the tumor proved then to be a benign ganglioneuroma.

To determine if our patient's tumor could have been similar to that described by Cushing and Wolbach, more special stains have been made recently. However, these stains give no cause for disagreement with the original diagnosis of fibrosarcoma.

On the most recent examinations in July 1955, the girl was healthy and showed the normal secondary sex characteristics of a girl nearly thirteen years of age; her menstrual periods had begun. There were no neurologic changes and no palpable masses. Her only complaint was occasional slight pain in her left hip when performing certain acrobatic dances.

The Spontaneous Cure of Massive Fibrosarcoma

Berner RE; Laub DL

Plastic and Reconstructive Surgery 36(2): Aug 1965; 257-262

Extracted Summary

A case of complete resolution of a massive congenital fibrosarcoma of the head and face is presented. No clue as to the reasons for such resolution was found. The unpredictable course of these poorly classified lesions is emphasized and should be kept in mind when a decision regarding therapy is made.

Selected Case Report

A female infant was born spontaneously after full term normal gestation. At birth the infant was lusty and vigorous, but presented a massive tumor involving the left side of the head, face and neck. The neoplasm extended from the parietal region of the skull downward almost to the shoulder, anteriorly to the midcheek and posteriorly to the occiput. An incisional biopsy was performed at age 10 days at another hospital. The microscopic picture of the lesion at that time was that of a juvenile fibroma, or fibrosarcoma, and the patient was subsequently transferred for further study.

When readmitted, at age 17 days, the infant was strong and well except for the very large mass over the left side of the head. The skin overlying the mass was integrately fixed and showed many dilated veins on the surface. The consistency was almost homogenous, of firm rubbery quality. There was no mobility in any portion and the deep component was firmly fixed to the deep structures of the neck and to the skull. The auditory canal passed directly through the mass and was almost three times normal length because of the tumor thickness. The canal passage was collapsed by the encroachment of the mass.

Surgical exploration was carried out through a long, preauricular incision extending from the temple down around the ear lobule and over the mastoid region. Three sizable segments of the tumor were excised for microscopic study. The gross surgical and frozen section histologic evaluation of the tumor revealed it to be highly invasive, without any potential planes of dissection about any aspects of the mass. There was no possibility of excising the tumor without massive mutilation. The incision was closed with the feeling that no effective surgical extirpation was possible and that the infant would not survive this growing lesion.

Pathologic examination by permanent section of the three resected areas revealed the same microscopic findings. The deep dermis of the overlying skin was replaced to a great extent by a well-differentiated collagen-forming tumor. The cells showed considerable uniformity of the nuclei, being darkly stained and spindle shaped. The cellular cytoplasm was scant and fairly eosinophilic. The cells were, for the most part, separated by dense collagen. Focal areas contained closely approximated nuclei, giving the appearance of giant cells. Mitoses were difficult to find. The tumor surrounded blood vessels and nerve sheaths and extended into the superficial dermis. The microscopic diagnosis was well-differentiated fibrosarcoma with massive collagen formation. The description is classical for fibrosarcoma but this could also be classified as a juvenile fibroma.

Two weeks after surgery, when the infant was 7 weeks old, the fixation of the skin overlying the tumor appeared to lessen and the tumor was softer. Within 4 weeks the size diminished and definite regression was visible. By 8 weeks, the mass reduced to one-half the original size and the overlying skin was normally movable. The mass disappeared at age 20 weeks, only 17 weeks after it had reached its maximum size. The child has been seen at frequent intervals and there has been no recurrence.

The course of resolution was dramatic and complete. Although prolonged follow up study must be continued, it is highly unlikely that there will be any recurrence. The patient has been carefully followed for over 3 years without any suggestion of return growth of the tumor.

Beneficial Effects of Immunotherapy (Bacterial Toxins) on Sarcoma of the Soft Tissues, Other than Lymphosarcoma

End Results in 186 Determinate Cases with Microscopic Confirmation of Diagnosis 49 Operable, 137 Inoperable

Nauts HC

Cancer Research Institute Monograph 16: 1975; 219 pgs

Extracted Summary

The purpose of this review is to analyze the effect of bacterial toxin therapy on patients with sarcoma of the soft tissues. It is hoped that the analysis of factors influencing success and failure in the following 186 cases will lead to more effective use of the method and to greater knowledge regarding this form of therapy and its effects, not only on sarcoma.

The treatment of malignant tumors by injections of bacterial products is based on approximately 400 recorded cases of so-called "spontaneous regressions," the great majority of which occurred in patients who concurrently developed an acute infection, principally streptococcal or staphylococcal. The present report consists of all the microscopically proven cases of operable and inoperable sarcoma of the soft tissues in which Coley toxins were administered.

Selected Case Report

Case 7: Very large recurrent inoperable fibrosarcoma of the gluteal region and thigh, confirmed by microscopic examination at Roosevelt Hospital, New York. Previous History: Mrs. M.S., female, aged 29, of Hartford, Connecticut. The family and early personal history were not recorded. Onset, in 1886 pain developed in the upper left thigh where the patient had been lying for some time while in bed following her third confinement. Three months later she noticed a growth the size of her hand a little above and posterior to the great trochanter. There was a constant ache in the leg. The growth gradually increased in size for four years. In October 1891 it had attained a diameter of 15 by 20 centimeters.

Surgery: It was then removed by Dr. Charles McBurney at Roosevelt Hospital in New York. The wound healed in six weeks.

Clinical Course: Three months later a recurrence developed further up in the gluteal region. This increased in size and caused a great deal of pain. The case was regarded as hopeless by McBurney. The patient was then referred to Dr. W.B. Coley. Physical examination at the time the toxins were begun showed a very large tumor occupying the left gluteal region, involving the muscles and fascia of the upper thigh, posteriorly. The tumor was so extensive that any attempt to remove it by further operation was out of the question. Pain was very severe.

Toxin Therapy (Type IV): Injections were begun by Coley at New York Postgraduate Hospital on April 7, 1894. They were made locally and the dose was gradually increased to the point of producing a marked reaction. The pain ceased but improvement was very slow.

On June 30, 1894 at the end of nearly three months' treatment the patient left the hospital. While there had been considerable decrease in size, there still remained a large tumor. This continued to decrease in size without further treatment.

Clinical Course: Examination by Coley five years later showed two small, hard movable masses at the site of the original tumor. On April 29, 1899, Coley operated at Memorial Hospital. The fibrous remains were separated from the surrounding tissue down to their connection with the bone, and this was cut through. Microscopic examination of the tissue showed it to be fibrous stroma, all the malignant cellular elements having been absorbed. The patient remained in good health and was examined periodically by Coley. She was presented before the Clinical Congress of Surgeons of North America in November 1912, and before the American College of

Surgeons in 1932. Her family physician, Dr. Philip G. McClellan, reported on October 10, 1939: "She is now 77 years old and at the site of the original lesions in the buttock there is nothing but a very pliable scarring. Her only symptoms are of a cardiac nature." She died on February 2, 1941, of coronary heart disease, at the age of 77. This was 51 years after onset and 47 years after toxin therapy.

Comment: The toxins caused regression of the malignant elements of the growth, but there remained a residue of hard fibrous tissue. This has been observed in a few other cases of fibrosarcoma, especially where one or more operations had been performed prior to toxin therapy. (Original reference: Coley WB; Glasgow Med J 126(2): Aug 1936; 49-86)

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Fibromatosis

Congenital Generalized Fibromatosis (Renal and Skeletal) with Complete Spontaneous Regression

Teng P; Warden MJ; Cohn WL
Journal of Pediatrics 62(5): May 1963; 748-753

Extracted Summary

A case of congenital generalized fibromatosis with involvement of the kidney and skeleton has been observed over an 18 month period. A total of 31 tumors in bone and one renal tumor was noted. These tumors were first found when the patient was one month old. The tumor in the kidney was surgically removed, and 2 skeletal tumors were biopsied. Complete spontaneous regression of the tumors in the bones was noted 16 months later.

Selected Case Report

The patient was the fourth child of the family. The prenatal period was uneventful, and he was born through a normal delivery on December 10, 1960. The birth weight was 5 pounds, 4 1/2 ounces. He appeared to be a normal baby. At 4 weeks of age, he developed projectile vomiting and was admitted to the hospital. A pyloric tumor was palpable, upper gastrointestinal series demonstrated an elongated pylorus, and a Ramstedt pyloromyotomy was performed. At the operation, the pylorus was hypertrophic, and there was no evidence of neoplastic involvement. However, intra-abdominal exploration disclosed a retroperitoneal mass involving the right kidney; no attempt was made to remove the tumor at this operation.

The postoperative course following pyloromyotomy was uneventful. Examination of the urine and blood chemical determinations, including blood urea nitrogen, calcium, and phosphate content, were noncontributory. On January 13, 1961, an intravenous pyelogram and retrograde pyelogram disclosed a distorted right ureter and deformed calyces of the right kidney. The pyelograms showed multiple lesions involving the pelvis and both femurs. A bone survey demonstrated multiple cystic lesions, some of which showed sclerotic margins in both diaphysis and metaphysis of both humeri, upper ulnas and femurs, upper tibias, pubes, and ischia. The lesions formed clusters in the metaphyses of the femurs, the right humerus, and the tibial bones. There were 2 areas of erosion the size of a peanut and pea, respectively, with sclerotic margins, in the left parietal region about 4 centimeters apart. A total of 31 bone lesions was counted.

On January 14, 1961, when the patient was 5 1/2 weeks old, a laparotomy was performed. A right transperitoneal approach was used to explore the right kidney, which was found to be approximately twice the normal size with a whitish gray tumor involving its lower half. There appeared to be no local spread of the tumor and there were no enlarged lymph nodes in the area. The excised kidney weighed 27 grams, measured 4.5 by 3 by 2.5 centimeters, and the tumor measured 2 by 3 by 2 centimeters. One week later the lesion in the distal left femur was biopsied. The lesions in the left parietal region were noted to be growing steadily in size. One and one half months later, the larger one reached the size of an olive. He was referred to the Neurosurgical Service where examination showed that the nodule was distinctly visible. The tumor was fixed to the cranium but the overlying scalp was freely moveable. X-rays of the skull showed the diameter of the larger defect measured 3 centimeters and that of the smaller one 0.8 centimeters.

On April 12, 1961, under local anesthesia, the larger cranial tumor was removed. The tumor was found firmly adherent to the outer layer of the dura but did not penetrate its inner layer. The smaller one was left intact for the purpose of observation of its outcome together with the rest of the bone lesions.

Gross pathologic examination showed all 3 tumors to be grayish white in color and in places faintly yellow. In some areas, the tumors were encapsulated, but in other areas there was no clear demarcation from their host tissues.

Microscopic examination revealed similar cellular structures in the renal, cranial, and femoral tumors. Their outstanding feature was proliferation of fibrous tissue. The cellular population consisted of maturing fibroblasts, uniformly spindle in appearance with oval or fusiform nuclei. The cytoplasm was moderately abundant, faintly acidophilic, and drawn out into long processes arranged in fascicular pattern with poorly defined boundaries. There were no giant tumor cells and mitotic figures were infrequent. The wall of the capillaries was lined by a single layer of endothelium and there was no evidence of endothelial proliferation. There were no bare axons noted in the lesions. Small areas of early degeneration were obvious

amid interlacing fasciculi. The renal tumor showed, distinctly, lack of encapsulation. At the periphery of the tumor, interlacing bundles of fibroblasts invaded and destroyed adjacent renal tissues.

The diagnosis of congenital generalized fibromatosis was kindly confirmed by Dr. Arthur P. Stout. The patient was discharged from the hospital on April 17, 1962, and closely followed at the Pediatric Clinic. He was noted to be growing satisfactorily. In August, 1962, a radiologic study showed that the cranial defect where the parietal fibroma had been removed was still present, but the small adjacent lesion, which had been left intact, had disappeared. There was no tumor palpable over the scalp. The multiple lesions of the long bones and pelvis had completely vanished. Recalcification of the bone lesions was complete. The long bones showed normal cortices and trabeculae. A few faint streaks of dense calcification at the metaphyses of the femurs are the only indication of the previous sites of the bone lesions.

The right diaphragm remains elevated as it has been prior to the pyloromyotomy and nephrectomy. Fluoroscopy with barium meal disclosed the stomach in normal position. The liver was not enlarged.

A bone survey and physical examination of the other 3 children of the family revealed no indication of bone pathology or evidence of fibromatosis.

Congenital Mesenchymal Tumors

Kauffman SL; Stout AP

Cancer 18(4): Apr 1965; 460-476

Extracted Summary

The authors' survey of 120 congenital mesenchymal tumors (37 cases of fibromatosis, 4 cases of fibrosarcoma, 15 cases of rhabdomyosarcoma, 17 cases of mesenchymoma, 13 cases of smooth muscle tumors, 11 cases of hemangiopericytoma, 2 cases of malignant hemangioendothelioma, 10 cases of histiocytic tumors, 3 cases of lipoblastic tumors, 2 cases of myxoma, and 6 cases of glomus tumors) emphasizes the rarity of truly malignant neoplasms of this type in the newborn. Certain tumors did not appear at all in malignant form, and other histologically malignant tumors behaved in a benign fashion.

The only types known to have metastasized were rhabdomyosarcoma, malignant mesenchymoma and fibrosarcoma. Among the benign tumors only the fibromatoses were associated with significant morbidity or mortality and this is because the fibrous tissue either grew diffusely destroying an extremity or formed masses in vital organs.

Forty tumors, about one-third of those surveyed, were histologically malignant. Nine of the 40 infants died of malignant tumor, 6 of these from rhabdomyosarcoma and one each of malignant mesenchymoma, fibrosarcoma and leiomyosarcoma of the prostate. There are too few cases available as yet for an understanding of the peculiarities of each type of congenital mesenchymal tumor. Our study emphasizes that congenital mesenchymal tumors rarely behave malignantly and suggests that congenital sarcomas differ biologically from those in the older age groups which they resemble histologically.

The authors present summaries of sixteen cases from their collection of 120 cases of congenital mesenchymal tumors.

Congenital Generalized Fibromatosis

A Case Limited to Osseous Lesions

Heiple KG; Perrin E; Aikawa M

Journal of Bone and Joint Surgery (United States) 54-A(3): April 1972; 663-669

Extracted Summary

A rare case of congenital generalized fibromatosis, apparently limited to the skeleton, has been described. A presenting fracture through a distal femoral lesion healed rapidly and uneventfully in Bryant's traction. The subsequent course was that of complete regression of all lesions within six to nine months.

Selected Case Report

A white boy, B. C., was born on September 10, 1967, and was first seen by us on January 17, 1968. He was born after a full-term uncomplicated pregnancy and delivery, and his birth weight was 2.89 kilograms. The neonatal period was normal and the child thrived. Subsequent inquiry as to maternal illness, use of drugs during pregnancy, or unusual environmental stresses was negative. The child was breast fed and later had normal diet and vitamin administration.

On January 17, 1968, the patient fell 60.9 centimeters from a couch and had immediate pain and deformity of the right femur. Roentgenograms revealed a fracture through a cystic lesion in the distal part of the right femur. Multiple similar lesions were discovered throughout the skeleton. The child was placed in Bryant's traction.

There were two older, normal siblings and a third who was stillborn in 1965 with deformities of unknown cause. The mother and father were both living and in normal health. Admission roentgenograms show the multiplicity of lesions. The following bones were involved: skull, six lesions (3 to 8 millimeters x 20 millimeters); maxilla and mandible, none visualized; cervical spine, lesions in centra of the second and fourth cervical vertebrae; thoracic spine, centrum of the third thoracic vertebra (double), centrum of the sixth thoracic vertebra (collapsed on left), centrum of seventh thoracic vertebra (collapsed on right), lesions in centra of the eighth through twelfth thoracic vertebrae; lumbar spine, probable lesions in all centra; sacrum, probably in the first sacral vertebra, possibly others not well visualized; pelvis, three to four 0.5 to 1.0 centimeter lesions in each ilium, several in pubic rami and ischia; ribs, lesions almost every rib at proximal end, also one to three lesions in the shafts of most lower ribs; clavicles, lesion at the inner end; scapulae, lesion in each scapular neck; humeri, lesions in the proximal and distal metaphyses but not in the proximal humeral epiphysis; radii, lesions in the proximal and distal ends; ulnae, lesions in the proximal and distal ends; wrists and hands, essentially normal, but a questionable lesion in right capitate and left thumb (proximal phalanx), metacarpals and phalanges otherwise normal; femora, lesions in proximal and distal ends, including both distal femoral epiphyses; tibiae and fibulae, lesions in both proximal and distal ends but not in the proximal tibial epiphyses; feet, lesions in each calcaneus and talus, none in cuboid, metatarsals, or phalanges. Over 100 separate bone lesions were easily identified.

Laboratory studies on admission were: total protein, 6.3 gm/100 ml (albumin 69%; globulin alpha 1-4%, alpha 2 11.5%, beta-11.5%, gamma-4%); blood urea nitrogen, 10.5 mg/100 ml; calcium, 10 mg/100 ml; phosphorus, 5.6 mg/100 ml; alkaline phosphatase, 8.6 Bodansky units; hemoglobin, 11.6 gm/100 ml; hematocrit, 32%. The blood sedimentation rate was 38 mm/hour. Blood culture was negative.

On the day following admission, January 18, 1968, a biopsy of the proximal lesion of the left tibia was performed under general anesthesia. The initial impression on review of the pathological sections was that the diagnosis was neurofibroma. This has been the common diagnosis in other reported cases of this disorder. However, the fracture healed rapidly and the case was thought to be clinically inconsistent with neurofibromatosis. After considerable discussion, it was decided to attempt to ablate the major lesions of the lower extremity to try to avoid major deformities that might occur if there were repeated fractures, as expected, through the obviously weakened segments. Therefore, on February 16, 1968, we curetted the lesions in the proximal and distal parts of the left femur and tibia and packed them with bank bone (frozen irradiated cancellous homograft). At surgery each involved metaphysis had a paper-thin, expanded shell of cortex and a cortical window was cut with a scalpel blade. The tissue of the lesion was firm and whitish-gray. It was not gritty and was curetted easily from the medullary space. There was none of the translucent quality associated with cartilage. Despite the roentgenographic appearance each metaphyseal lesion was uniloculated. Culture was negative. A double hip spica cast was applied and the patient was discharged on February 20, 1968. Readmission was planned for surgery to the opposite lower extremity in two months.

At readmission on April 4, 1968, the cast was removed and roentgenograms were made. To our astonishment, all the lesions, not just those which had been removed, showed marked regression. Because of this, surgery was cancelled and it was decided to follow the patient without further active treatment. The child continued to be perfectly healthy clinically and the bone lesions continued to regress rapidly over six to nine months and by the age of twenty-two months only faint bone scars remained to indicate the site of the original lesions.

At the time of last examination in the spring of 1971, when the child was just under four years of age, he had neither clinical nor roentgenographic evidence of the prior process except for slight irregularity of the collapsed vertebral bodies (the sixth and seventh thoracic vertebrae). He is of normal height and weight for his age and has had a normal male sibling born subsequently.

Congenital Generalized Fibromatosis with Complete Spontaneous Regression

Schaffzin EA; Chung SMK; Kaye R

Journal of Bone and Joint Surgery (United States) 54-A(3): April 1972; 657-662

Extracted Summary

A case of congenital generalized fibromatosis with multiple bone involvement is reported. The disease is often fatal, and often involves many systems with fibroblastic lesions which are present at birth or shortly thereafter. Our case is the fourth reported case with extensive bone involvement. All of the lesions in our case have completely or almost completely disappeared during the first eighteen months of life.

Selected Case Report

The patient, a white girl, was born at term weighing 2.5 kilograms. The circumstances of gestation and delivery were normal. There was no family history of neurofibromatosis. At birth the patient had fifty-nine firm subcutaneous nodules, 0.25 to 1.5 centimeters in diameter, distributed over the head, trunk, periosteum, and upper and lower extremities with the greatest concentration over the back.

A subcutaneous nodule was removed and the initial diagnosis was neurofibromatosis. The infant fed and sucked poorly and frequently regurgitated and she had a weak cry. She gradually improved but continued to gag and regurgitate. She was admitted at the age of six weeks to The Children's Hospital of Philadelphia, because of coughing during and after feedings, and respiration stridor, especially after feeding. Her appetite was good and she had reached her developmental milestones adequately. Her general physical examination, except for rhonchi, noted on both inspiration and expiration, was normal. There were multiple firm nodules under the skin which ranged from fully moveable to relatively fixed, including one over the right scapula and one over the medial part of the left clavicle. A cavus deformity of the right foot was present with complete absence of function of the anterior tibial and extensor hallucis longus muscles. This was felt to be due to pressure of a fibrous lesion on either the sciatic, anterior tibial, or peroneal nerve.

The routine laboratory studies were normal. The roentgenogram of the lungs was normal, as was an electroencephalogram. The roentgenograms showed multiple lytic lesions of the ribs, left clavicle, femora, tibiae, fibulae, and humeri. There were no lesions in the skull. A number of the soft tissue nodules contained ill-defined areas of calcification. In comparison with roentgenograms made during the first few days of life the clavicular lesion appeared to be resolving, but new lesions had appeared in the distal parts of the femora and in proximal parts of the tibial and fibular shafts. A severe gastroesophageal reflux was demonstrated and direct laryngoscopy showed paralysis of the right vocal cord, but no nodules could be seen involving the larynx. The slides of the initial biopsy specimen were reviewed and the diagnosis was changed to congenital fibrosarcoma; subsequently, it was again changed to generalized fibromatosis. Due to incoordination of the swallowing mechanism, a gastrostomy and jejunostomy were performed on the seventeenth day after admission. The patient tolerated tube feedings well and except for an episode of obstruction of the pylorus by the gastrostomy tube requiring repositioning of the jejunostomy, there were no subsequent nutritional problems and the patient was discharged from Children's Hospital at age five months doing well. The right foot was supported in a bivalved leg cast and stretching exercises were begun. Over the next few months she partially regained function of the paralyzed muscles of her right leg and electromyograms of the involved muscles of the right lower extremity done at age six months were normal.

Many of the subcutaneous lesions disappeared during the first year of life and the remainder decreased in size. Roentgenograms made at thirteen months of age showed complete healing of the majority of the bone lesions and partial healing of the remainder. At the last follow-up, when the patient was eighteen months old, virtually all nodules had resolved and the patient's swallowing function, as shown roentgenographically, was without evidence of aspiration. It was planned to remove the gastrostomy tube.

Recurring Digital Fibroma of Infancy

Bloem JJ; Vuzevski VD; Huffstadt AJC
Journal of Bone and Joint Surgery (Great Britain) 56-B(4): Nov 1974; 746-751

Extracted Summary

Three typical cases of recurring digital fibroma of infancy are reported, with a follow-up of three to four years. In each case excision of the tumours during the first year of life was followed by recurrence and then by some degree of slow spontaneous resolution, in one case complete. A strictly conservative approach is recommended for three reasons: the difficulty of complete excision, the tendency to spontaneous regression and the facts that no case of metastasis and no case of persistence into adult life have yet been reported. Cytoplasmic inclusion bodies could not be demonstrated in the biopsy material from these cases, nor any virus.

Selected Case Report

Case 1: A boy aged six months was seen in 1969 because of a swelling on the back of the distal part of the right middle finger. He was the fourth child of healthy parents. The tumour had appeared in the third month of life; it was about 5 millimeters in diameter, firm, not tender, and fixed to the skin but not to the underlying tissues. Biopsy showed a dermatofibroma with no sign of malignancy. The tumour then stared to grow more rapidly, and it was decided to excise it completely and apply a skin graft. Three months later a recurrence was excised. After three more months a second recurrence had occurred and the parents refused further operations. Now after four years this had disappeared and only a small local thickening is evident on the lateral aspect of the finger where the graft meets normal skin.

Myofibroblastic Contraction in Spontaneous Regression of Multiple Congenital Mesenchymal Hamartomas

Benjamin SP; Mercer RD; Hawk WA
Cancer 40(5): Nov 1977; 2343-2352

Extracted Summary

Subcutaneous nodules from a newborn boy with multiple fibromatosis involving the head, neck, trunk, and all four extremities were studied by light microscopy, transmission electron microscopy, and immunofluorescent techniques. Light microscopy suggested a hamartomatous process with fibroblastic, adipose, vasoformative and apparent smooth muscle components. The principal cell population combined ultrastructural characteristics of both fibroblasts and smooth muscle cells. Immunofluorescent studies revealed binding of human anti-smooth muscle antibody to the cytoplasm of the spindle cell population of the subdermal nodules but not to fibroblasts of the overlying uninvolved skin. The ultrastructural and immunofluorescent studies revealed the previously undescribed fact that fibrous hamartoma of infancy is principally a proliferation of myofibroblasts. At age 8 months, there was complete spontaneous regression of all subcutaneous nodules not previously altered by excisional biopsy. The authors conclude that myofibroblasts are fibrocontractile cells, which play a role in shrinkage and eventual disappearance of these subdermal hamartomas.

Selected Case Report

The patient, a 5-day-old white male infant, was admitted to the Cleveland Clinic Hospital for evaluation of multiple (24) subcutaneous nodules involving the right post-auricular scalp, right mandible, left shoulder, right and left upper arm, forearms, upper and lower back, lower anterior chest, abdomen, buttocks, hips, thighs, and lower legs. The skin nodules were noted immediately following birth. The infant's mother enjoyed excellent health during her pregnancy and related no history of infection, trauma, or drug ingestion. There was no family history of skin tumors. On physical examination the subcutaneous nodules varied from 0.5 to 6 centimeters

in greatest dimension and were uniformly elevated, indurated, freely moveable, and apparently nontender. There was no discoloration of the overlying skin. Radiographic examination demonstrated erosion defects in the midshaft of the right humerus, distal right femur, tibia, fibula, and right fifth rib. Excisional biopsy of a 1 centimeter nodule on the left shoulder was performed. Macroscopically, an unencapsulated irregular, gray-white, firm dermal mass was noted to infiltrate the subcutaneous fat. Tissue was submitted for light microscopy, electron microscopy, and immunofluorescent studies. The patient was discharged on no specific therapeutic regimen.

At age 1 year, all of the nodules had disappeared with the single exception of a 1 centimeter lesion over the left greater trochanter which was excised and submitted for similar studies. At age 15 months, there was no evidence of persistent or recurrent disease.

A Case of Infantile Digital Fibromatosis Showing Spontaneous Regression

Ishii N; Matsui K; Ichiyama S; Takahashi Y; Nakajima H
British Journal of Dermatology 121(1): Jul 1989; 129-133

Extracted Summary

A case of infantile digital fibromatosis (IDF) in a 3-year-old girl is reported. Two tumours were present on the left fourth toe and one over the lateral aspect of the left heel. Histological examination revealed that the tumor cells contained intracytoplasmic inclusion bodies, characteristic for this condition. The tumours showed spontaneous regression without therapy.

Selected Case Report

The patient, a 3-year-old girl, was referred to the dermatology clinic, Yokohama City University Hospital, in June 1986. She had multiple small nodules on the left foot which were firm, smooth and painless. Two were on the left fourth toe and measured 0.8 centimeters and 0.1 centimeters in diameter, respectively, and another was on the lateral aspect of the left heel, being 1 centimeter in diameter. A biopsy of the lesion on the heel was performed and histologically, the tumour was composed of elongated, spindle-shaped fibroblasts arranged in whorls or interdigitating sheets, with abundant collagen bundles. Faintly eosinophilic intracytoplasmic spherical inclusion bodies were seen on staining with haematoxylin-eosin. These also stained bright red with Masson's trichrome and purple with phosphotungstic acid haematoxylin (PTAH), and yellow with van Gieson stain.

Immunohistochemistry for vimentin, desmin and keratin was performed using the peroxidase antiperoxidase complex (PAP) method. Positive staining for vimentin was noticed in the cytoplasm of the tumour cells, whereas desmin and keratin were negative.

All three tumours grew slowly after biopsy. However, in December 1986, regression of the tumours became apparent and by June 1988 the tumours were only small macules.

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Congenital Generalized Fibromatosis with Predominant Osseous Involvement in a Chinese Newborn
Chan YF; Lau JHK; Tong CY
Journal of Pediatric Orthopedics 9(1): 1989; 64-68

Desmoid Tumors

Desmoid Tumors Particularly as Related to Their Surgical Removal

Strode JE

Annals of Surgery 139(3): March 1954; 335-340

Extracted Summary

Desmoid tumors are slow growing and do not metastasize, but they may ultimately undergo sarcomatous degeneration. They are curable by complete removal, but continue to grow when operation is incomplete. They should be removed early so as to prevent the necessity of sacrificing a large part of the abdominal wall. However, even though as much as half of the anterior abdominal wall must be removed to effect a cure, the method described, using fascia lata to replace this structure, has been found most effective. The surrounding parietal peritoneum lends itself to extensive mobilization and for this reason large defects can be completely peritonealized. Attention is called to the regression of a desmoid tumor concomitant with onset of menstruation. No similar case has been found reported in the literature and the significance of this observation has not been determined. This case and three others are outlined in the article.

Selected Case Report

The second case, a Chinese female, was first seen on November 3, 1938, at the age of seven, with an extensive tumor of the abdominal wall. The tumor had been first noted as a small nodule arising in the suprapubic area when the child was one year of age. On November 18, 1938, an attempt was made to remove the tumor surgically, but it had invaded the abdominal wall so extensively that this could not be accomplished. A large amount of the tumor was resected followed by irradiation, but little of value was accomplished.

On July 30, 1943, resection of a portion of the tumor was again done, followed by irradiation with the hope of palliation. By November 24, 1943, the tumor had enlarged considerably, and, at that time, it was our belief that probably the lesion was undergoing sarcomatous degeneration and that life expectancy was correspondingly short. The tumor became ulcerated and the patient was confined to bed for a period of six months.

The patient was lost sight of until recently, when, much to our surprise, it was learned that she was not only living, but was well, had been at work for the past five years, and was contemplating marriage. The physician, under whose care she has been for the past ten years, stated that nothing specific had been used; no roentgen ray or radium treatment in particular.

The patient's father ascribes the improvement in part to Chinese herbs, the nature of which has not been determined. Examination of the patient's abdomen reveals areas of thickening and induration of the abdominal wall, in all probability due to residual areas of tumor, though there has been a marked regression in the tumor since 1943. The patient states that the tumor is slowly becoming smaller; there is no ulceration.

Since clinical improvement began about the time of onset of menstruation, it would seem logical to conclude that in all probability ovarian function played a part in the patient's marked improvement. It is interesting to speculate what effect pregnancy might have on this tumor, and also the ultimate cessation of ovarian function.

Desmoid Tumours

A Series of 33 Cases

Dahn I; Jonsson N; Lundh G
Acta Chirurgica Scandinavica 126: 1963; 305-314

Extracted Summary

The aetiology, localization, and treatment of desmoid tumours are considered. Heavy muscular contractions (such as at childbirth) seem to be a most important aetiologic factor. Hormonal factors may secondarily influence the growth of the desmoid tumour.

From the standpoint of therapy, it is important to draw attention to the fact that desmoid tumours may occur extra-abdominally. Treatment consists in microscopically radical excision. Of 24 radically excised desmoid tumours, only two recurred. Cortisone was used without success in one case. Radiation therapy failed to produce any effect in another instance.

Attention is called to Gardner's syndrome; hereditary intestinal polyposis with fibroid tumours (including desmoids), osteomas, and epidermoid cysts. One case is presented of colonic polyposis with excessive desmoid infiltration in the abdominal scars.

Selected Case Report

Case 4: Female, 40 years, nulliparous. Case record 2149/52, Department of Surgery, Kalmar. A tumour of slow growth had been present in the right side of the groin for six months. In 1952 a hard tumour, the size of an orange moveable in relation to the underlying structures, was palpated in the right iliac fossa. A biopsy specimen showed it to be a desmoid tumour. No further measures were undertaken. On examination eight years later, the patient reported that since taking the specimen the tumour had gradually decreased in size and had wholly disappeared after two years. Menstruation ceased at the same time.

Spontaneous Regression of a Desmoid Tumour

Jenkins NH; Freedman LS; McKibbin B
Journal of Bone and Joint Surgery (Great Britain) 68-B(5): Nov 1986; 780-781

Extracted Summary

A 15-year-old girl presented with a very large Desmoid tumour in her buttock; it extended into the pelvis and thigh and would have required hindquarter amputation for its removal. This was not performed and the tumour underwent spontaneous regression. Fourteen years later the patient is alive and well. The suggestion is made that in some of these cases a more expectant approach to treatment might be justified for what is essentially a non-malignant condition.

Selected Case Report

The patient, a 15-year-old girl, presented with a history of several months' left-sided sciatica; her pain was constant but unrelated to movement or activity. Examination revealed a large mass filling the left buttock, arising from within the musculature and extending anteriorly to the adductor compartment of the thigh and inferiorly to the mid-thigh level. There was also a mass 6 x 8 centimeters in the left iliac fossa which on rectal examination seemed to be continuous with the mass in the buttock through the greater sciatic notch. This was subsequently confirmed by retroperitoneal air insufflation. Movements of the hip joint were extremely limited. There were no lesions elsewhere in the body and a chest radiograph was clear. A barium enema, intravenous pyelography and sigmoidoscopy were also normal.

A biopsy was performed from the most distal part of the tumour at the back of thigh. It was there found to be arising from the musculature, principally from the gluteus maximus and the hamstrings; the sciatic nerve was totally surrounded by tumour. Subsequent histological examination of the material removed revealed an infiltrating mass of fibrous tissue with degeneration of the adjacent muscle. There was no evidence of malignant change and the tumour was considered to be a benign musculo-aponeurotic fibroma (Desmoid tumour).

The patient was seen by a number of surgeons whose unanimous decision was that adequate local excision was impossible without irretrievable damage to the limb, and that a hindquarter amputation would therefore be necessary. This was considered but, in view of the patient's

age and the fact that the condition was non-malignant, it was decided to continue observation and to treat her symptoms.

However, the main mass continued to grow, reaching its maximum size two years after her initial presentation. At this stage there was a huge swelling of the buttock which was clearly visible through the patient's clothing and the hip had become fixed in a position of flexion and medial rotation. Satellite lesions had developed behind the left knee and severe flexion contracture of this joint developed. By then, however, the patient's sciatica had largely subsided. At this stage elongation of the hamstring tendons was performed to provide a satisfactory correction of the knee flexion deformity. Despite these events, the patient remained active and uncomplaining and successfully took her matriculation examinations.

About five years after the initial presentation the tumour began to regress spontaneously, initially from the adductor group. The communication between the pelvic and buttock masses resolved and the hip became mobile. By 10 years after its appearance the tumour was no longer palpable in the lower limb; the pelvic mass also diminished considerably in size, although it was still just palpable.

Seven years after the initial presentation a painful lump had developed in the left calf. Clinically this appeared to be a lesion similar to that in the buttock and no biopsy was undertaken. It expanded over the following year and gave rise to an equinus deformity of the ankle which was treated by elongation of the tendo calcaneus. Soon afterwards this mass also underwent spontaneous regression and disappeared completely six years after it was first noticed.

At review 14 years after the development of the initial tumour the patient remains in good health with no palpable mass in the thigh and only a small mass in the pelvis as previously described. There are no residual contractures in the hip which has a full range of movement, the knee is fully mobile and there is no residual equinus deformity. Despite the residual pelvic mass, the patient has recently had a vaginal delivery of her first child.