

Adamantinomas of the Jaw.

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Adamantinomas are usually stated to be fairly rare tumours. When H. G. B. ROBINSON 1939 reviewed the literature he found 379 cases. At The Norwegian Radium Hospital, Oslo, there have in the period from 1931 to 1948 been treated a total number of 10 cases. A review of this material is of some interest.

The adamantinomas are derived from epithelium that has the power to differentiate into enamel epithelium without actually forming enamel. It is true that BERNAYS (1885) and CHIBRET (1894) reported two cases in which it was found; but none of the later investigators have been able to demonstrate enamel in the adamantinomas. K. THOMA in his "Oral Pathology" proposes the term "Adamantoblastoma" as it describes the histologic make-up much better.

The tumour may form from cells that have already become differentiated or from embryonic epithelial cells. In the latter instance the result will vary according to the stage in which the cells differentiate during the tumour development. It must be remembered that no matter from what structure the tumour has formed it inherits the potentialities of the embryonic oral epithelium, and that, therefore, acini of glands occasionally may be found in the tumour (K. THOMA).

The tumour may be derived from:

1. Genetic disturbance of the enamel organ or the tissue forming it.
2. Cell-rests of the enamel organ.

3. The epithelium of odontogenic cysts (BECKER 1930 and CAHN 1933).

4. The surface epithelium covering the jaw (BAKAY 1909, KAUFMANN 1922, GULLIFER 1936).

5. Displaced epithelium in other parts of the body.

CHRITCHLEY and IRONSIDE, 1926, PEET 1927 gave an account of adamantinomas in the hypophysis, ZAWELOWSCHIN reported a case of adamantinoma in the ovarium in 1931, FEINBRUN 1930 reported a case in the pharynx and FISCHER a case in the tibia.

The adamantinomas are distributed fairly equally between the sexes.

The summary of ROBINSON's findings after his study (1937) of the 379 cases reported in the literature is given in Table 1.

Table I.

	Cases
Total number of cases	379
Sex (311 cases)	
Male 45.7 %	142
Female 54.3 %	169
Average age at time for report 37.6 yr.	248
Average duration of tumour 8.5 yr.	232
Average age at time for discovery 30.1 yr.	222
Site of growth (293 cases)	
Mandible 83.7 %	247
Maxilla 16.3 %	48
Structural characteristics (219 cases)	
Cystic 57.5 %	124
Cystic and solid 24.2 %	53
Solid 19.1 %	42
Contained unerupted teeth	14
Enamel pearls, keratinization	16
Recurrence after operation	119
Metastases or histologic evidence of malignancy 4.5 %	

The adamantinomas are usually found in the lower jaw in the region of the premolars and molars, the ramus and the symphysis of the mandible. In the maxilla they are less frequent. Here the maxillary sinus generally becomes involved with expansion of both the facial and nasal walls. Its growth is very slow and usually the soft tissue is not involved with the exception of a few atypical cases. The malignant tumours such as the carcinomas, on the other hand both invade and destroy tissue without consideration.

The age incidence varies greatly, the average being about 30 years.

The tumour exists often a long time before the patient has

any knowledge of it, giving in the beginning no symptoms. The adamantinomas are usually a central tumour but occasionally they are of periferic origin (GULLIFER 1936). In some cases the tumour is contained in a single compartment, when it simulates an odontogenic cyst. It may even contain a tooth so that the diagnosis of a dentigerous cyst seems logical, and the mistake generally is found out only at the operation.

Teeth are quite frequently found in adamantinomas. The tumour may be formed round the crown: Dentigerous adamantinomas, at the side of the tooth: Periodontal adamantinomas or they may be formed from several enamel organs. HILDEBRAND described a case containing about 200 teeth, in other words, an odontoadamantinoma.

Unusually they are of a polycystic type and the new cysts are formed in the neighbourhood by invading epithelium. The tumour may cause a swelling of the alveolus or of the entire jaw. The compacta may be destroyed and then a tumour is formed that is fluctuant but not painful on palpation. But when infection occurs it will be painful.

The question of malignancy is a matter of great importance. Recurrence in these tumours is very common and is due to the fact that because the tumour grows slowly the surgeon is tempted to be too conservative and the patient so late gets any therapy. RHEINMÖLLER (1937) states that a fairly radical excision when the tumour is small prevents such recurrence and more serious resections. LANGE (1936) endorses AXHAUSSEN's recommendations of a biopsy examination before deciding the treatment. In this way it may soon be discovered malignant tendencies in both the tumour epithelium and the stroma. LEONE MAC GREGOR has reviewed the atypical and more or less malignant cases reported in the literature, a total number of 28 cases (1935, ROBINSON 1937 only accepting 8) MAC GREGOR found the following atypical microscopic features in these tumours:

1. A marked cellularity of the stroma, in some cases as extensive as in fibrosarcoma, or
2. A predominance of the cuboidal over the stellate and cylindrical types of epithelial cells, or
3. A preponderance of epithelium over stroma with marked intertwining and treelike branching of the epithelial processes.

As to recurrence, it must be kept in mind that conservative treatment with incomplete removal tends to disrupt the expansive

growth of the tumour and induces a more malignant and infiltrating type. SIMMONDS (1928) believes that conservative operation might be advised for young people with small tumours, if the patient agrees to frequent examination and subsequent radical operation if there is recurrence. The recurrence is due to proliferation of epithelium in the spongiosa in the neighbourhood. Where the tumour-cells are less differentiated they show here as otherwise a more infiltrative character. Local metastases occur very rarely (SPRING 1932 reported such a case). When it occurs it might be due to the adamantoblastic epithelium or to sarcomatous changes in the stroma. Such tumours may be looked at as adamantosarcoma.

As to the metastases to the lymph glands they occur very rarely and must be verified by microscopic examination. SIMMONDS (1928) has reported such a case. VORZIGER and PERPLA (1932) has reported a case of adamantinoma with inhalation metastases to the lungs. As a rule the adamantinomas are benign and the growth is very slow. But it must be kept in mind that a slowly growing tumour may become malignant and grow more rapidly (KAUFMANN 1922, MAYER 1929, RISAK 1927).

As to the roentgen findings the monocystic type is not always easy to differentiate from an odontogenic cyst. But a notch in the cyst wall or very small accessory cysts in the adjoining bone are significant. Teeth are often found in both. But the tumour encloses the tooth competely in the tissue, while the cyst usually displaces the adjoining teeth. A further sign of tumour is complete denudation of the roots of a tooth. As to the polycystic types they are usually more easily recognized. But a central giant-cell tumour and a central myxofibrom may give a picture like this.

But the expansion is here slighter than in advanced adamantinomas, and in the myxofibroma there is trabeculation with tendency to form lines through the tumour instead of round compartments. In adamantinomas the cysts are generally shown as small circular shadows. The differentiation between a solid tumour and a monocystic type is not possible without an operation. The possibility exists that the solid type may become cystic or the cystic type may become solid by ingrowth of tumour tissue from the wall. The solid tumour is usually white in color, of fine granular consistency and encapsulated. In the cystic type we find some compartments filled with solid tumour and

others which are cavities lined by a smooth membrane consisting of epithelium backed by a small amount of connective tissue. In such cysts there is a clear, yellow fluid, sometimes of mucous, sometimes of colloid character. The periost is generally not involved.

By microscopic examination there are found great variations.

The solid tumour consists of a branching network of solid epithelial strings composed of cuboid-cylindrical cells in a connective-tissue stroma, or the branching epithelial strings may be composed of a network of stellate cells surrounded by a palisade-work arrangement of cylindric cells, perhaps with an intermediate zone of more cuboid, polygonal or oval cells between these two layers. The histologic components of the cystic adamantinoma are the same, but here a cystic transformation has taken place, notably in the layer of stellate cells. There is great difference of opinion as to the development of these cysts. Probably they are formed by vacuolar degeneration of the epithelial cells. KROMPECHER emphasizes that it may be due to absorption of colloid into the epithelial cells. PAPADAMITRIOU is of the opinion that the fluid also may be formed by actual secretion from the cells of the enamel pulp.

Characteristic for the adamantinoma are the stellate cells, in connection with the palisade cells. The stellate cells have a central nucleus and a long protoplasmic process while the cylindric cells have an oval, slightly stainable nucleus lying towards the interior of the cell string.

But the structure may be very different from this typical picture. Often the cells are cuboid or polygonal, in one or several layers or they may be more irregularly disposed. But usually there will be some epithelial string or island that shows a more typical picture and gives the diagnosis.

Author's Material.

At The Norwegian Radium Hospital, Oslo there have in the period from 1931 to 1948 been treated 10 cases of adamantinomas, 7 female and 3 male. The distribution according to age etc. is given in Table II.

In all cases but one the tumour was situated in the lower jaw, about equally distributed on each half. In the majority of cases the tumour was situated around angulus mandibulae, in

Table II.

Distribution acc. to age:	10—20 yr. 1 case	20—30 yr. 1 case	40—50 yr. 2 cases	50—60 yr. 4 cases	60—70 yr. 2 cases
Duration of tum. before consulting a physician:	5 weeks 1	2 months 1	5 months 1	2 years 2	4 y. 8—10 y. 1 1
Duration of tum. altogether:	4 years 1	5 6 7 9 11 20 y. 33 y. 1 1 3 1 1 1 1			

ramus ascendens or ramus horizontalis, only in one case towards the median line on the left side. Only in one case the tumour was localised to the maxilla.

A remaining tooth in connection with the tumour was found in two of the cases. In most cases, the symptom which brought the patient to a doctor was a swelling at the site of the tumour, in one case tumour + pains. Trismus appeared later in one case, pain in another case.

The diagnosis was verified by biopsy in 7 cases. In the remaining cases the roentgenological findings were found so characteristic that the diagnosis of an adamantinoma seemed justified. The further control too has confirmed the diagnosis in two of these cases. In one case the patient was admitted with the diagnosis of a malignant tumour in the parotis. But the roentgen examination revealed a destruction in the lower jaw, and the diagnosis of an adamantinoma was verified four years later, the biopsy then showing the picture of an adamantinoma.

In a second case, which is described a little more fully later and where the diagnosis of adamantinoma was made when the disease had existed for a period of four years, the tumour became malignant and the patient died five years later after the first treatment here. In two cases operation was out of the question owing to the widespread infiltration of the tumour. Five cases had undergone only one operation. One of these cases was operated upon after the treatment at The Norwegian Radium Hospital, resection of one half of the jaw and transplantation of bone being done. One case had been operated upon five times altogether and one case ten times, the last time while a patient here. As to the treatment The Norwegian Radium Hospital is of the opinion that the effect of the X-ray therapy has been greater in some

cases than is generally realized. Of course an operation must be performed as soon as possible in those cases, where there is any chance for the complete removal of the tumour, without destroying the continuity of the bone. In advanced cases this is out of the question and then X-ray therapy ought to be tried. Naturally, there will never be a *restitutio ad integrum*. The swelling of the bone will persist but the tissue in the tumour may be destroyed and the growth of the tumour arrested. Most of the cases have been treated by teleradium, in some instances combined with roentgen treatment. In the cases where the infiltration of the tumour has been far advanced, roentgen treatment has been given over greater areas than is possible by teleradium. At the Norwegian Radium Hospital the dosage generally given is the same as for malignant tumours. Up to four series of treatments were given in one case, but the dosage varied somewhat.

Treatment with teleradium:

In 2 cases, one course was given with 3,350 r. units and 4,500 r.

In 2 cases, two courses were given with 4,350 r. and 1,530 + 1,020 r. units through two portals. In the second case 2,776 r. and 3,280 r. units.

In one case three courses were given with 4,670, 3,570 and 2,550 r.

In one case teleradium was given with 3,800 r. + roentgen therapy 1,000 + 1,000 r.

In one case teleradium was given with 3,280 r. and in succeeding treatments radium-mould with 3,327 r. units (the physical data for the teleradium treatment being: 3,000 mg. Rad. E. target-skin distance 6.2—7.2 cm. filter 1 mm. platina + 3.3 mm. brass; for the roentgen treatment: 175 Kv. 4 ma., target-skin distance 40—50 cm. filter 0.5—1—2 mm. Cu + 0.5—3.5 mm. Alm.)

The courses of the disease in these cases are given in Table III.

Table III.

Died	Cured	Free from symptoms or station. process	Destruction with pseudarthrosis	Recessed lower jaw with bone transplantation
1	3	4	1	1

As to the two cases which were successfully treated, an operation was performed at an early stage, the tumour infiltration not being far advanced. In both cases postoperative X-ray treatment was given. One of these cases was a boy, 18 years of age. The operation resulted in the removal of a solid tumour. In the third case the disease was cured by X-ray treatment.

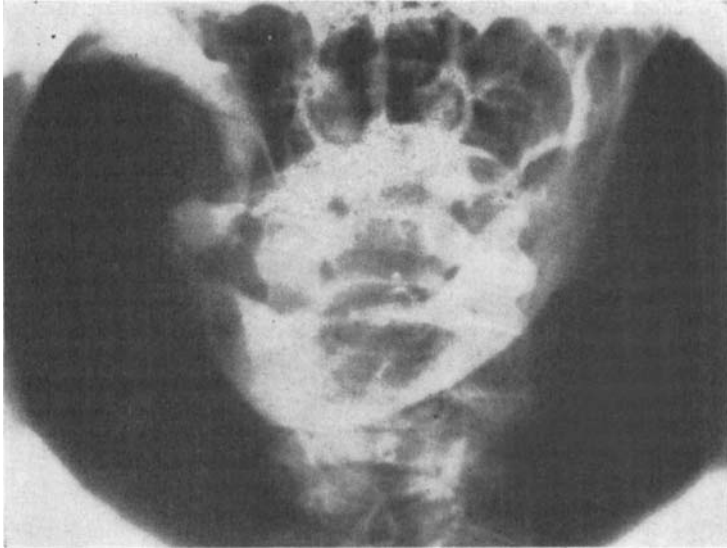


Fig. I. Shows the great defect and destruction in the mandible.

The tumour in the case which received the most intensive X-ray treatment on the other hand was only slightly affected.

The diagnosis of an adamantinoma may be difficult, the three cases without histological verification plainly showing this. In two of these cases infection with accumulation of pus was found, and this might mask the presence of epithelial cells. Furthermore one must consider the fact that when the disease has existed over a long period, perhaps with varying treatments, some degenerative processes may have supervened, and then the active tissue of an adamantinoma may hardly be found without a radical operation.

Three cases which are of special interest must be described a little more fully:

Case I. Woman, 56 years old (J. nr. 265/37) had been operated upon thirty years ago. A tumour in front of the right ear was removed. It was supposed to be a mixed tumour of the salivary gland type, but a great part of the mandible on the right side had to be removed. After thirty years the tumour recurred (July 1936), growing at first rather slowly. The patient was admitted to The Norwegian Radium Hospital, Oslo in April 1937. A great, ovoid tumour was found, extending from the retromandibular region on the right side towards the corner of the mouth. The surface was found rather plain and the con-

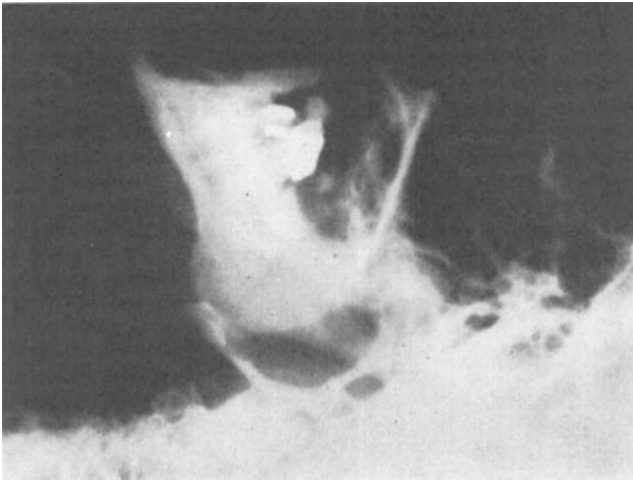


Fig. II. On the right side.

sistency firm. No fluctuation and no tenderness by pressure was found. The mucosa inside the cheek on the right side was found thickened. Roentgen examination (Fig. I) disclosed a great defect in the mandible on the right side after the operation and a tumour involving a great part of the right ramus ascendens of the mandible. Teleradium treatment was given with 4,370 r. without any effect. In the beginning of 1943 the tumour again started growing. She was readmitted to The Radium Hospital, Oslo, April 7th 1943. The tumour was found extending from the right processus mastoideus to the right corner of the mouth and from the cheek bone to the border of the lower jaw. It was of a rather firm consistency, fluctuation being found in some small areas. Teleradium treatment was again instituted with 3,570 r. through three portals. July 1944 a new tumour appeared, a little distance beneath the other tumour. She was readmitted, September 11th 1944. Puncture of the tumour was done with evacuation of a deep-red humor. Teleradium was again given with 3,060 r. The tumour still went on growing and roentgen therapy was then instituted with 2,100 r. through two portals. A perforation developed growing into a rather big defect. A biopsy showed: Fibrous granulation tissue. Roentgen examination January, 14th. 1948 shows (Fig. II): A great destruction in the right half of the lower jaw. Biopsy, January 1, 1948 has verified the diagnosis of an adamantinoma (Fig. III).

The histological picture showed that the tumour was of a somewhat atypical appearance with great cords and strands of atypical epithelial cells, bounded by a margin of epithelial cells resembling basal cells. As previously mentioned it must be kept in mind that conservative treatment with incomplete removal tends to disrupt the expansive growth of the tumour and induces a more malignant and infiltrating

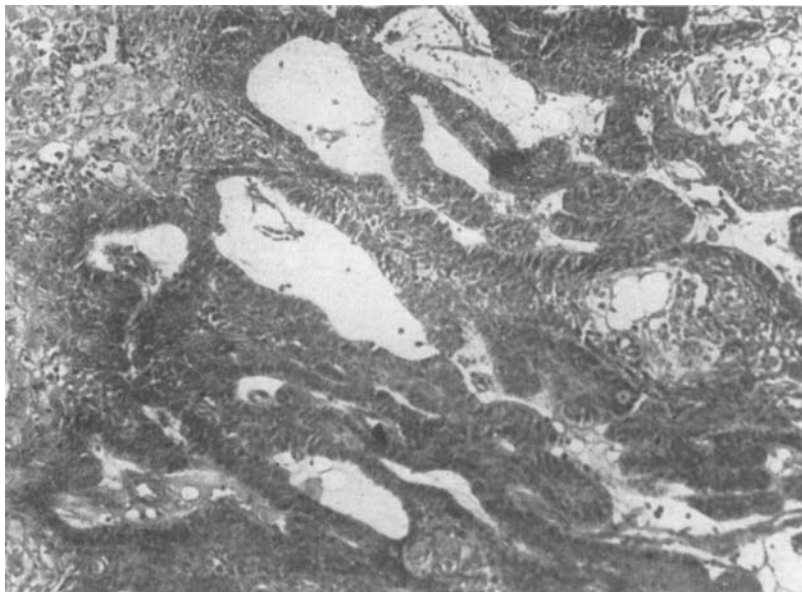


Fig. III. Shows the great cords and strands of epithelial cells, bounded by a margin of epithelial cells resembling basal cells.

type. In this case some atypical microscopic feature was found but no evidence of malignancy was proved. A peculiar feature was that the tumour recurred after so many years.

Case II: Man, 51 years old (J. nr. 543/43), who in 1938 had "a gum-boil" in the lower jaw on the left side. After a year it perforated into the mouth. July, 1942 the tumourous swelling reappeared in the same place, growing slowly, giving pains and trismus. He was admitted to the Hospital of Drammen, x-ray examination showing: Adamantinoma mandibulae. The tumour perforated spontaneously, the perforation being enlarged by incision. After repeated incisions he was subjected to x-ray therapy, operative therapy not being indicated. He was then admitted to The Radium Hospital, Oslo, June 16th 1943. Here a great swelling was found, comprising the whole cheek region on the left side, from the temple region to the upper part of the neck. Located in the angle of the lower jaw was a tumour 5×7 cm. in size, the surface being uneven and irregularly bounded. The skin over the tumour was deeply bluish in color and some fistulae were found. The tumour was of a rather firm consistency and adherent to the lower jaw. The skin was not adherent.

By palpation from the mouth, the alveolar process on the left side was involved in the tumourous swelling from the angulus to the premolar region. The x-ray picture showed: A great destruction in the lower jaw on the left side. D: Adamantinoma.

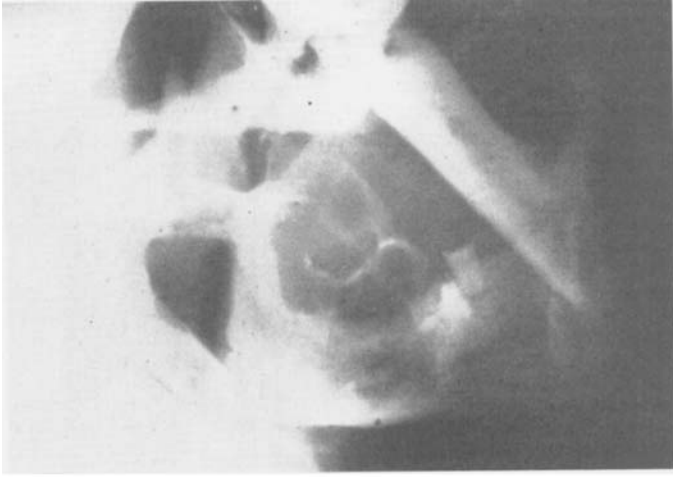


Fig. IV.

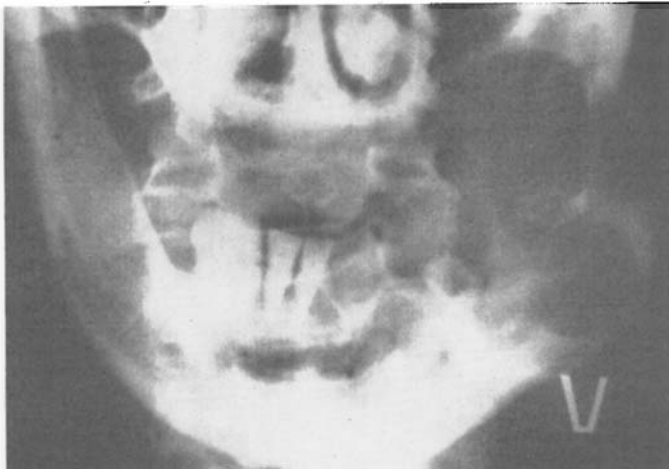


Fig. V. Shows a polycystic adamantinoma in the mandible on the left side (Man, 51 years old).

As a characteristic feature round compartments and cavities lined by irregularly formed trabeculation were found. (Fig. IV.)

The histological examination showed, May 20th, 1943: Papillomatous epithelial growth with some atypical structure and inflammation.

X-ray therapy was given with $3,700 + 3,600$ r. through two portals.

After some temporary decrease the tumour again started growing.

At the control, January 20th, 1944 a considerable progress of the tumours was found with ulcerations, infiltrating the skin, the subjective symptoms being correspondingly increased.

During the following years the tumour grew rapidly, displaying several profuse bleedings. He died, November 23th, 1945.

It is regrettable that it was not possible to obtain a positive biopsy in this case. X-ray examination by two x-ray centers shows features



Fig. VI. Reveals a great tumour in the maxilla on the left side (Man, 67 years of age).

characteristic of an adamantinoma. By the clinical course of the disease it must be assumed that the tumour has become malignant.

Accordingly the case must be regarded as belonging to the registered cases of adamantinomas, malignancy probably being induced, but not histologically verified.

Case III: Man, 67 years old (J. nr. 827/37). Two of his sisters and brothers had died of cancer ventriculi. For some years he had complained of indigestion, when ulcer duodeni was proved and he had undergone medical treatments twice. About three months previous to the admission in November 8th, 1937 a molar in the maxilla on the left side became loose and on drawing the tooth a perforation appeared, communicating with the sinus maxillaris and the cavum nasi on the left side.

Examination on admission to this hospital revealed a great tumour in the maxilla on the left side, comprising the alveolar process and a great part of the palate on this side. Corresponding to the drawn tooth a defect, the size of an almond was found communicating with the sinus maxillaris. (Fig. VI).

An enlarged submaxillary lymph node the size of a bean was found on the left side.

Roentgen examination revealed blurring of the left maxillary sinus and a defect in the bone in the molar region perforating into the sinus maxillaris.

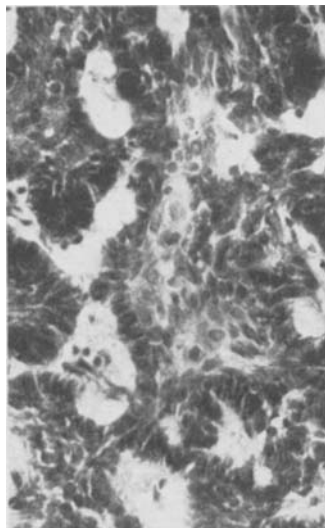


Fig. VII. Microphotography reveals the appearance of a solid adamantinoma (Man, 67 years of age).

Microscopy of biopsy sample, November 12th, 1937 resulted in the diagnosis of: Carcinoma with adamantinoid structure.

Teleradium was given with 4,480 + 4,560 r. through two portals.

The patient was readmitted in March 8th, 1938 and roentgen therapy with 1,200 + 1,200 r. units was given. Some cleaning of the ulcer had then occurred, but still a communication between this and the sinus was found. Roentgen examination revealed the tumour to have decreased in size. In June 6th, 1938 radium-therapy was again instituted, a mould being applied into the sinus through the perforation in the bone. 2,870 r. units were given. Further control has revealed the tumour to have decreased in addition and no recurrence was proved.

The patient died in December 13th, 1946, succumbing to intercurrent diseases (bronchitis and atherosclerosis).

The case was recorded and treated as a carcinoma. A control of the

biopsy sample from November 12th, 1937 reveals evidence of an adamantinoma. (Fig. VII.)

Microscopy (R. EKER): The tumour tissue generally reveals the appearance of rather densely packed, irregular chords and strands of epithelial cells. Some treelike branching and intertwining of the epithelial processes is evident. Generally the branching epithelial strings are composed of a network of slightly differentiated, stellate cells surrounded by a palisade-work arrangement of cylindric cells. Some atypical signs are found, the nuclei varying somewhat as to the size and chromatin-substance. A few mitosis is seen. The stroma is of edematous appearance and in some sections a considerable infiltration with lymphocytes, plasma-cells and myelocytes is seen. D: Adamantinoma.

The course of the disease in this case too has verified the diagnosis, the adamantinoma being cured only by x-ray therapy and no operation being performed.

Summary.

After reviewing the location, morphologic and histologic appearance of the adamantinomas, the pathogenesis and ethiologic relationship between this tumour and the enamel organ, and the therapy, the author gives an account of 10 cases of the disease treated at The Norwegian Radium Hospital, Oslo during the period 1931 to 1948. 7 of the patients were women, three were men. In all the cases but one, the tumour was situated in the lower jaw. 8 of the tumours were cystic, 2 solid. The treatment was chiefly roentgenologic, the most of the patients being operated previously to the admission or the process so far advanced that no radical operation was found possible. Doses were given as for malignant tumours. The Norwegian Radium Hospital is of the opinion that x-ray therapy is of some effect. Anyhow it can stop the progress of the tumour for some time.

Zusammenfassung.

Nach einer Übersicht der Lokalisation, des morphologischen und histologischen Bildes des Adamantinoms, der Pathogenese und ätiologischen Verwandtschaft dieses Tumors mit dem Emailorgan; und der Therapie, berichtet der Verf. über 10 Fälle dieser Krankheit, die in der Zeitspanne 1931—1948 in der Radiumzentrale in Oslo behandelt wurden. Sieben von den Kranken waren Frauen und drei waren Männer. In 9 Fällen sass der Tumor im Unterkiefer, nur in einer Falle im Oberkiefer. 8 der Tumoren

waren zystisch, zwei solide. Die Behandlung war hauptsächlich röntgenologisch. Die meisten von den Kranken waren früher operiert worden oder die Infiltration war so weit vorgerückt dass keine Operation indiziert war. Bei dem Norwegischen Radium-Hospital, Oslo, ist die Meinung vorherrschend dass die Röntgenbehandlung wirksam sei. Die Progression des Tumors will jedenfalls gehemmt werden.

Résumé.

L'auteur passe en revue la localisation ainsi que l'aspect morphologique et histologique des adamantinomes, leur pathogénie, leur relation étiologique avec l'organe de l'émail, enfin leur thérapeutique, puis il relate 10 cas de cette affection qui furent traités à l'Hôpital Norvégien du Radium, à Oslo, durant la période allant de 1931 à 1948. Sept des malades étaient des femmes, trois des hommes. Dans tous les cas sauf un, le néoplasme était localisé à la mâchoire inférieure. Huit des tumeurs étaient kystiques, deux solides. Le traitement a été principalement radiologique, la plupart des patients ayant été opérés avant leur admission, ou se trouvant atteints de lésions si avancées qu'aucune intervention radicale ne fut jugée possible. Les doses administrées furent celles des tumeurs malignes. A l'Hôpital Norvégien du Radium on estime que la radiothérapie est d'un certain effet. En tout cas elle peut arrêter pour quelque temps les progrès du néoplasme.

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