PERIOSTITIS DEFORMANS DUE TO WINE FLUOROSIS

by

M. Soriano
Barcelona, Spain

In 1952,I (1) described a new bone disease, which I called periostitis deformans. In 1965 (2) I was able to identify this disease with chronic intake of F due to habitual drinking of wine containing fluorine. I termed it "Wine Fluorosis".

This kind of chronic fluoride intoxication is rare. It differs materially from skeletal fluorosis. During a 15 year period we observed 29 cases. Fluoride was originally added to wines to retard fermentation in contravention of existing health laws. The F concentration in wines ranged between 8 and 72 mg/l (ppm). Large-scale consumption of such wines in certain areas has permitted us to study the bone changes thus produced. The most typical form is the development of periostitis deformans. In only one case did we encounter a purely osteomalacic pelvic form. Biopsy of the coxal bone revealed quantities of fluorine greater than 4000 ppm (ash basis). Table I presents the F analyses of bone in 6 patients.

TABLE I

F Content of Bones in 6 Cases of Wine Fluorosis

Case No.	Bone	PPM F	Consistency of Bone
4	Pelvic bone	11,800	Very dense
	Tibia (cortex)	4,500	Very hard
	Femur (cortex)	3,809	Very hard
	Tibia (spongiosa)	11,200	Very soft
	Femur (spongiosa)	11,900	Very soft
5	Femur (cortex)	905	Very dense
	Pelvic bone	9,770	Very porotic
	Femur	8,090	Spongy
	Humerus	2,880	Dense cortex
	Pelvic bone	8,504	Very soft
	Rib	7,320	Very soft
	Radius	7,690	Soft
6	Femur (cortex)	1,537	Very hard
	Tibia (cortex)	1,216	Very hard
	Tibia (head)	1,549	Slightly hard
	Trochanter	2,937	Slightly hard
11	Newly formed bone	11,750	Reticular, spongy
	Marrow	2,376	Hard
12	Osteophyte (rib)	11,700	Slightly hard
	Osteophyte (finger)	13,580	Soft
	Osteophyte (elbow)	11,400	Soft
	Pelvic bone	10,900	Hard
	Tibia	5,519	Very hard
13	Pelvic bone	13,400	Soft

Chairman, Faculty of Medicine, University of Barcelona, Spain.

SORIANO

Pathology in Periostitis Deformans

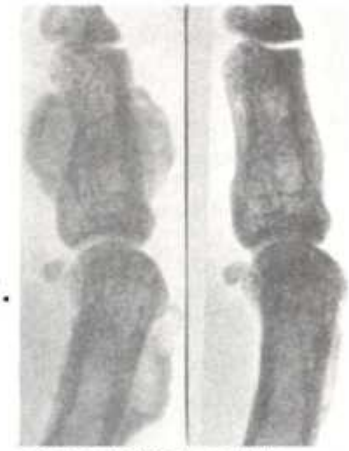
Four typical phases of F induced periostitis deformans were noted:

1. Endosteal changes lead to a condition which we termed sclero-atrophic osteitis. An initial, slowly developing osteosclerosis gradually leads to osteoporosis and bone atrophy.

2. Periosteal changes develop in the course of repeated episodes of hyperostotic periostitis deformans. During these outbreaks the affected bones exhibit zones of periosteal hyperostotis which grow prodigiously, until they resemble bone tumors. On the fingers, they reach the size of an almond (Fig. 1). On forearms and femur they can grow as large as an apple (osteoblastic stage Fig. 2). Usually after a period of about three to five months when the lesions cease growing, the osteoclastic stage (Fig. 3) sets in, and their size decreases (atrophic stage). If the lesions fail to clear up entirely, they leave permanent deformities in the affected bones (Fig. 4 and 5). The periosteal lesions are typical of the disease.

Fig. 1

(a) (b)



a) Periosteal nodule on finger.
b) Same finger 2½ years later in a state of remission.

3. Invading osteophytes cause soft-tissue changes in periostitis deformans, a condition which we termed "invading osteophytosis." Osteophytes develop in tendons, fasciae, and muscles at the areas where they are attached to bone. This condition can become widespread. Like the other hyperostotic changes observed in the disease, the osseous lamellae of osteophytes undergo atrophy and become osteoporotic during the late stage of the illness, although they do not decrease in size.

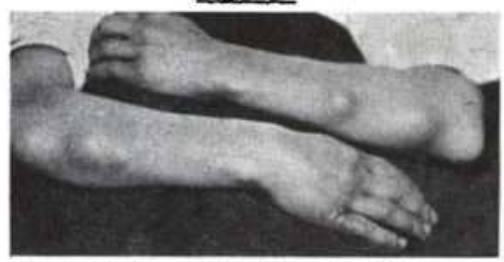
4. Joint changes or fluoric arthrosis may be very severe especially in the hip, knee and elbow joints (Fig. 6).

Radiological Diagnosis

In each group of periostitis deformans, the x-ray changes are characteristic.

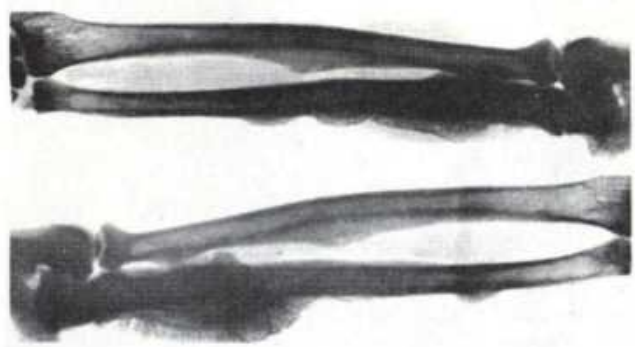
1. Sclero-atrophic Osteitis: At the onset of the disease osteosclerosis occurs in the vertebral column, the pelvis, and the shafts of the long bones. Subsequently osteoporosis develops at these original sites. The osteoporotic zones tend to expand toward the adjacent areas and eventually affect the cortex and the whole pelvis. In the final stage, osteoporosis invades the vertebral column and all remaining bones (Fig. 7,8,9.10).

Fig. 2 *



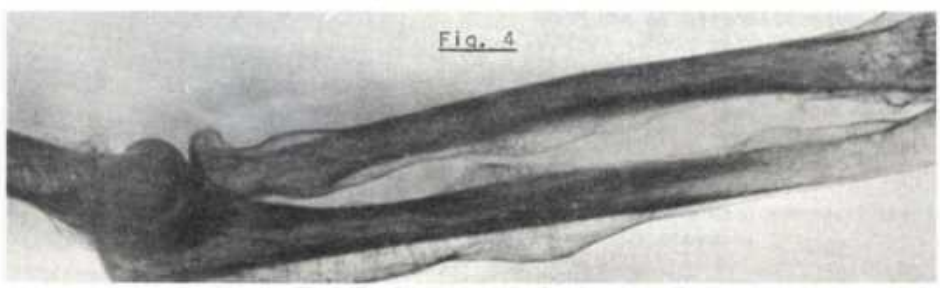
Periosteal ossification simulating bone tumors.

Fig. 3



a. Osteoblastic phase of periosteal growth in forearms in two stages of development.
b. Lamellar appearance of osteophyte. Deformity of both bones 1 year later

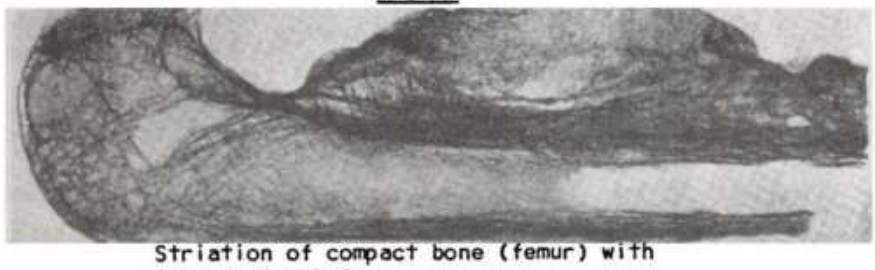
Fig. 4



Advanced periosteal growth in forearm (osteoclastic phase) in another patient. Osteosclerosis in diaphysis.

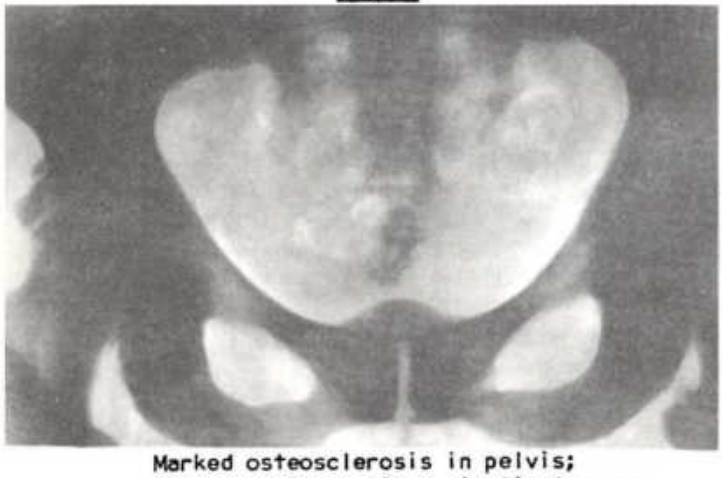
* All illustrations except Fig. 2 represent radiographs.

Fig. 5



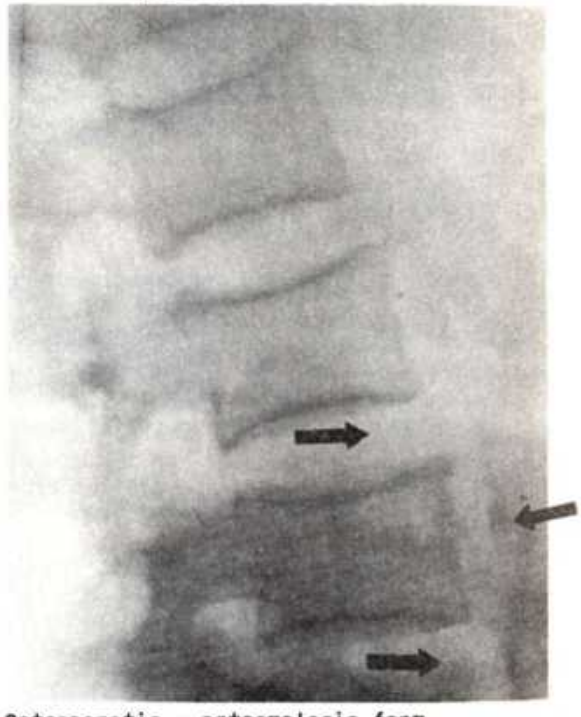
Striation of compact bone (femur) with large osteophyte.

Fig. 6



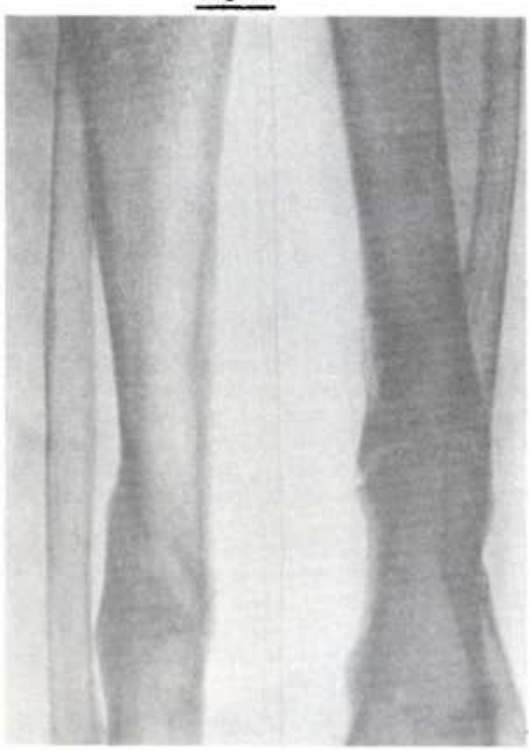
Marked osteosclerosis in pelvis; Intense osteophytic periarticular proliferation of hip joints;"Blocking" arthrosis. "Feathery" outline of os ischium.

Fig. 7



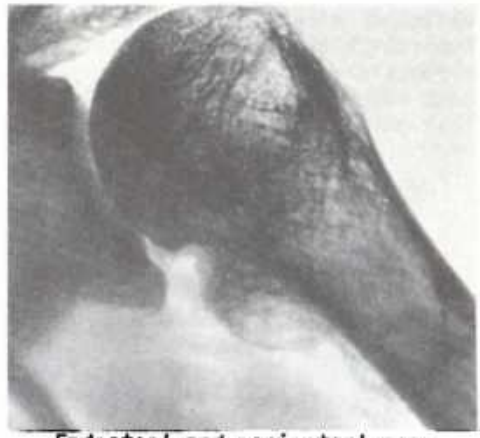
Osteoporotic - osteomalacic form of wine fluorosis. Homogenous softening of vertebral bodies. Calcifications in aortic walls.

Fig. 8



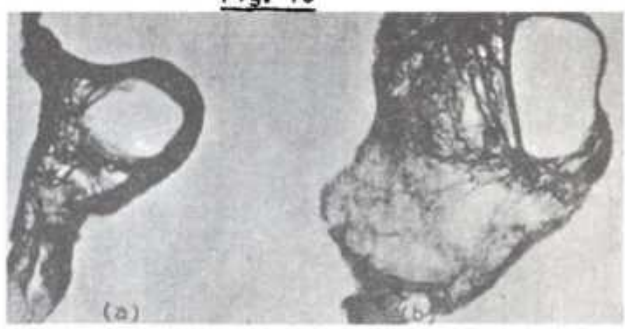
Radiograph of lower leg in wine fluorosis. Osteoporotic - osteomalacic form. (F content of coxal bone 8950 ppm, of wine 15.29 ppm.) Healing spontaneous fractures.

Fig. 9



Endosteal and periosteal proliferations of humerus. Marked osteoporosis in epiphysis. Thickened trabeculae; sclerotic cortex.

Fig. 10



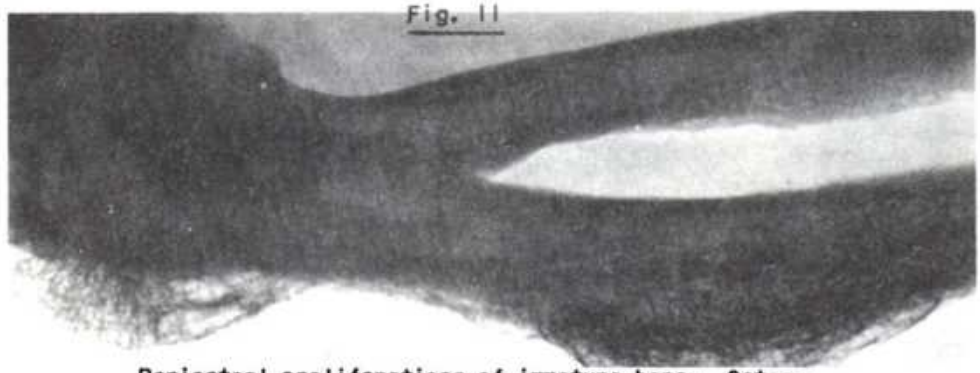
Cross sections of distal part of femur at epiphysis showing two stages: The early endosteal lesion with thickened cortex (a) and subsequent thinning and "insuflating" of bone tumor (b).

Areas of intense sclerosis coexist with those of marked osteoporosis in the same bone. This is highly characteristic of the disease.

2. Periosteal changes: During the early growing or osteoblastic period, pseudotumors appear which originate in the periosteum. They are composed of projecting nodules of newly formed bone demarcated by round or multicircular margins, which present a radial structure in the shape of a fan or cockade (Fig. 11,12). During the osteoclastic stage they assume the form of a fine

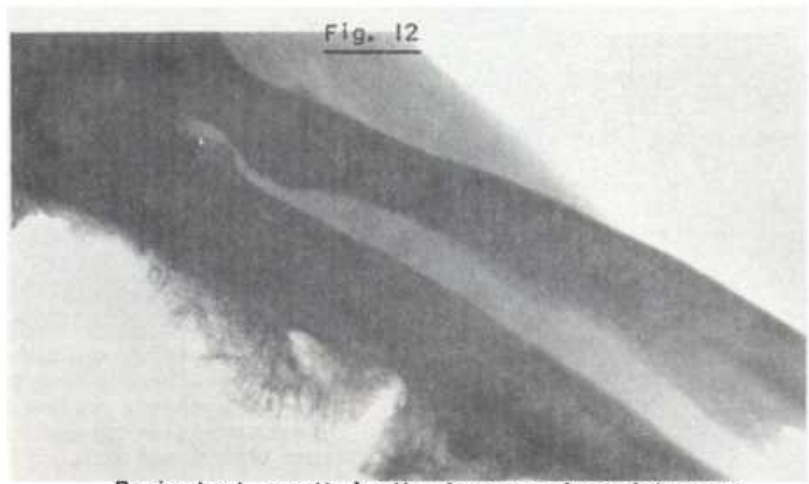
reticular structure which subsequently merges into progressive osteoporosis. Eventually the osseous lamellae become atrophic and appear honey-combed in the x-ray picture. When new areas of periostitis develop on top of the earlier ones, each layer of the newly formed bone corresponds to a new outbreak of activity. They go through the same stages as do the previously described lesions.

Fig. 11



Periosteal proliferations of immature bone. Osteoclastic phase (forearm).

Fig. 12



Periosteal growth in the forearms (pseudotumors) involving the interosseus membrane (invading osteophytosis). Osteosclerotic cortices.

During subacute outbreaks endosteal bone may also be invaded. The osseous lamellae of the compact endosteum will undergo an expansive outburst of activity and produce a bone tumor. The osseous lamellae develop quickly, thicken, separate from each other and leave wide spaces between them (Fig. 10). Later, the lamellae of the endosteal tumor undergo atrophy and form large pseudotumors in the endosteal bone. Frequently, the endosteal bulges are covered by periosteal bone structure which becomes intimately fused with them.

3. Invading osteophytosis: Osteophytes of endosteal or periosteal origin may proliferate and acquire bizarre forms. The osseous lamellae with irregular margins at the interosseous membrane of the forearm (Fig. 12) are typical of wine fluorosis. Equally as characteristic are the harpoon-shaped changes in the tibia and the rostrum-like osteophytes in the calcaneum. There may also be large thin plates which expand deeply into muscles. Occasionally, isolated bony nodules are seen within the sheaths of tendons and muscles which present the radiological feature of fibrositis and of myositis ossificans (Fig. 14). Calcifications in arterial walls are also frequently observed (Fig. 7).

4. Fluoric Arthropathies: Around joints, thick marginal osteophytes develop. In some instances, they grow to such an extent as to block joint movements ("blocking arthrosis") (Fig. 6). The joint block can also be induced by calcification of the periarticular ligament. The most common sites of articular involvement are the hips, the sacroiliac, elbow and knee joints. In older persons, the vertebral column is commonly affected. Advanced stages of the disease show atrophy and ulceration of joint cartilage. All changes of periostitis deformans tend to be symmetrical.

Latent Forms

At the onset of the disease, the vertebral column and the pelvis show only insignificant osteosclerosis together with a slight periosteal reaction and minor porotic lesions in the shafts of the long bones. The periosteal lesions regress eventually, leaving only thin porotic layers of periosteal bone. The most conspicuous finding in such patients is sclero-atrophic osteitis with invading osteophytosis and arthritic changes.

The areas of atrophic osteitis are likely to become the sites of spontaneous fractures. Osseous deformities remain, some of which are produced by residual hyperostoses of periosteal or endosteal origin. Less frequent are curvatures of the diaphyses (Fig. 3). New subacute episodes of periostitis deformans can occur at any stage of the disease.

The radiologist should suggest the fluoric origin of the disease and emphasize the need to search for the toxic substance in the patient's environment, i.e. the habitual intake of contaminated wine.

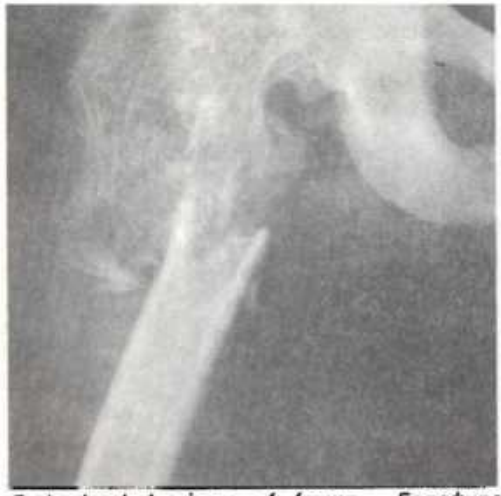
In order to confirm the diagnosis, a fluorine bioassay should be performed on a biopsied bone specimen taken from the iliac crest.

Fluoride Action on Bone Tissue

Fluoride stimulates osteogenesis. This causes the initial osteosclerotic stage as well as periosteal and endosteal reactions of proliferating bone tissue which resemble bone tumors. If the irritating action of F is intense and persistent the early osteoblastic reaction is followed by osteoporosis and bone atrophy. The osteoporotic stage of the disease occurs only when fluoric intoxication is very severe.

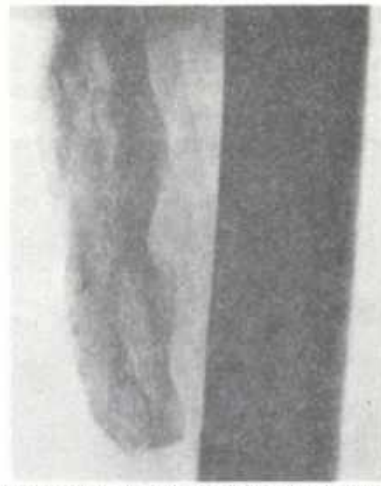
A most striking feature of the osteoblastic action of F is the fact that it can also result in osteoblastic metaplasia in tissue other than bone and cause fibrositis and myositis ossificans. Fibrositis ossificans is usually localized

Fig. 13



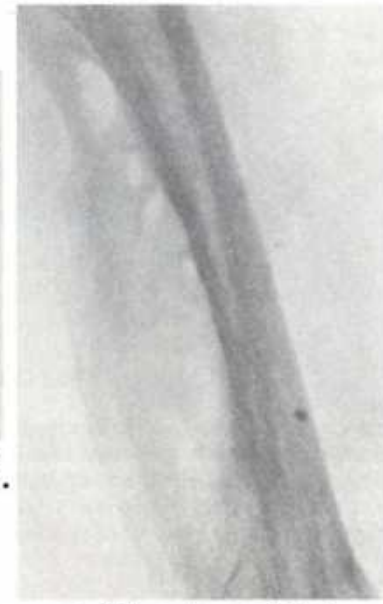
Endosteal lesions of femur. Fracture in an osteoporotic zone of the striated compact bone; atrophy of femoral head; osteosclerosis of pelvis.

Fig. 14



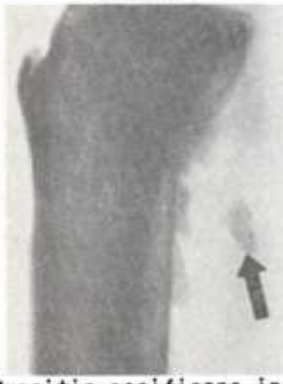
Ossification of muscle of thigh (myositis ossificans) with marked osteosclerosis of femur.

Fig. 16



Orifices in ossified musculature due to arteries passing thru them.

Fig. 15



Myositis ossificans in "Wine Fluorosis"(knee).

Fig. 17

Terminal case of periostitis deformans with invading osteophytosis; atrophy of head of femur. Osteoporosis alternating with osteosclerosis.

near the affected bone, but is nevertheless completely independent of it. The extension of invading osteophytosis into soft tissue encountered in periostitis deformans, appears to be due not only to endosteal and periosteal proliferation, but to a progressive fibrositis ossificans of membranes, tendons and fasciae which are attached at the sites of proliferating osteophyte activity. This "invasive osteophytosis" is another characteristic feature of the disease.

According to our observations the prolific growth in periostitis deformans continues as long as daily amounts greater than 8 to 10 mg of F are ingested no matter through what vehicle. If F intake is intermittent subacute outbreaks will occur coincidentally with increased F ingestion.

The development of the lesions is influenced by such predisposing constitutional factors as osteogenesis imperfecta and probably by impaired nutrition. In "wine fluorosis", alcohol contributes to the severity of the fluorosis, since chronic alcholism induces anorexia, gastritis and liver damage and thus leads to malnutrition.

Summary

On the basis of 29 cases observed during 1948 to 1968, the author reports a disease termed periostitis deformans which was caused in alcoholics by sodium fluoride added to wine in concentrations of the order of 8 to 72 ppm. Four different phases of the disease are described which are associated with osteosclerosis and osteoporosis. They lead to marked disability and may terminate fatality.

Bibliography

1. Soriano, M.: Madrid, Ed. Paz Montalvo 1952.
2. Soriano, M.: Revista Clin. Espan 97:375-388, 1965.