Section of Orthopædics

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Osteoid-Osteoma

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IT is about twenty years since the concept of osteoid-osteoma of bone began to shape itself in my mind. The idea was stimulated by the study of 5 cases observed in 1933. These were all cases in which there was a lesion in a spongy bone area with the clinical diagnosis of "localized osteomyelitis" or "bone abscess", but in which the tissue submitted for microscopy failed to show evidence of inflammation. Instead, it showed as the significant pathologic feature a nidus-like focus of osteoid and new bone whose removal promptly and permanently abolished the patient's complaints.

Search of the literature (including a number of radiographic atlases) yielded largely negative results. Feeling increasingly that the condition represented something distinctive, I gathered these 5 cases together for publication, formulating the name of "osteoid-osteoma" to characterize the lesion's pathological pattern (Jaffe, 1935).

It was not until 1938 that we realized that an osteoid-osteoma may also develop in relation to the cortex of a bone. This came to light in cases, involving long bones in particular, which were ascribed clinically to "sclerosing non-suppurative osteomyelitis" and "osteomyelitis with cortical bone abscess". Previously we had been puzzled by such cases because of the difficulty of reconciling the clinical diagnosis with the absence of microscopical evidence of inflammation in the bone shavings.

When the next pertinent case presented itself clinically, the surgeon was prevailed upon to do a block resection of the affected area, so that we could study the supposed abscess intact in its setting of thickened cortex. To my surprise, the area supposedly representing the cortical abscess turned out, on histologic examination, to represent the same lesion which, in relation to spongy bone areas, I had designated as osteoid-osteoma. When, through the study of additional cases, it became clear that such cortical lesions likewise belong in the category of osteoid-osteoma, the concept was fully launched (Jaffe and Lichtenstein, 1940). However, the general acceptance of osteoid-osteoma as a clinico-pathologic entity was rather slow, though, sooner or later, various centres have come to accord it recognition.

The osteoid-osteoma lesion.—The osteoid-osteoma lesion evolves slowly, and often gives rise to clinical complaints for many months before its presence is demonstrable in the X-ray picture at all, and even longer before the X-ray picture is reasonably clear-cut. The evolving lesion has two components: (1) the core or nidus-like focus (the osteoid-osteoma proper), and (2) the reactive perifocal zone of bone thickening.

The nidus may lie within the spongiosa, perhaps close to an articular surface, or may be associated with the cortex, and may lie against the inner surface of the cortex, within it, or even, though rarely, between the cortex and the periosteum.

The reactive perifocal zone is much more likely to be striking when the osteoid-osteoma is cortical than when it is within the spongiosa. In the latter case, one may find merely a zone of thickened, condensed, and otherwise altered spongiosa surrounding the nidus as a ring or band of variable extent. Thickening about a cortical nidus may extend over a considerable part of its circumference, especially when the lesion is in a long bone, which, furthermore, may be thickened for a number of inches longitudinally.

The nidus itself is usually roundish or oval, and is not more than 1 cm. in its greatest diameter. The clarity with which it shows up in the X-ray picture is affected by such variants as the position of the nidus in the affected bone; the radiodensity of the nidus itself; the extent and radiodensity of the perinidal zone of reaction; and the attention given to technique such as taking the picture from various angles, "coning down", overexposure, &c.

Against this general background, I shall now mention some cases illustrating various locations and various radiopathologic appearances of the osteoid-osteoma lesion.

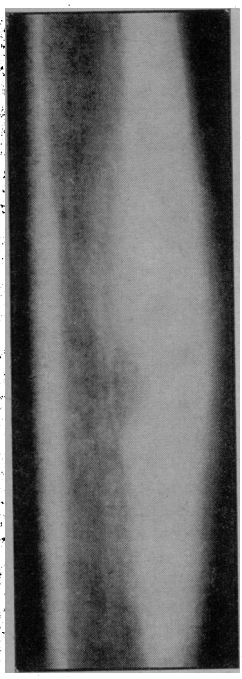
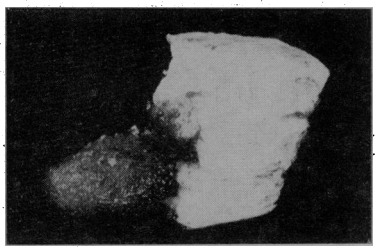


FIG. 1.



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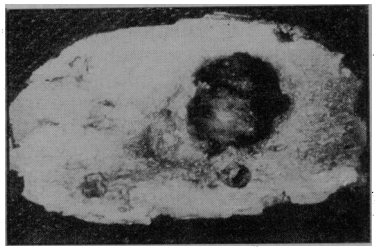

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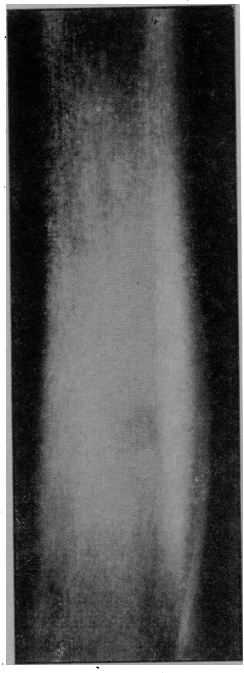

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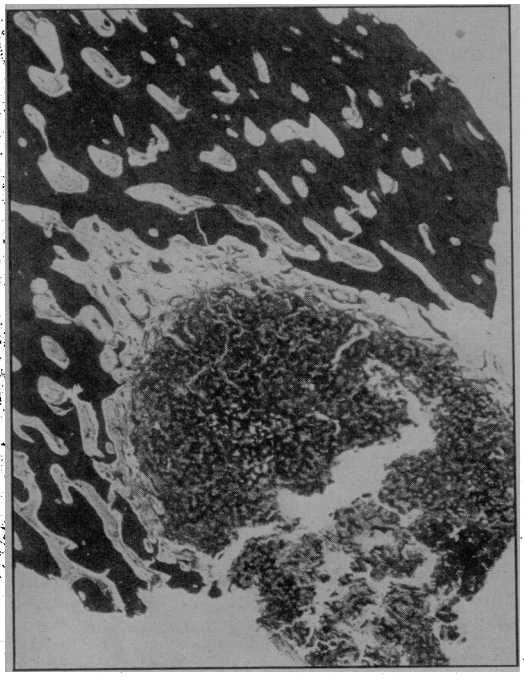

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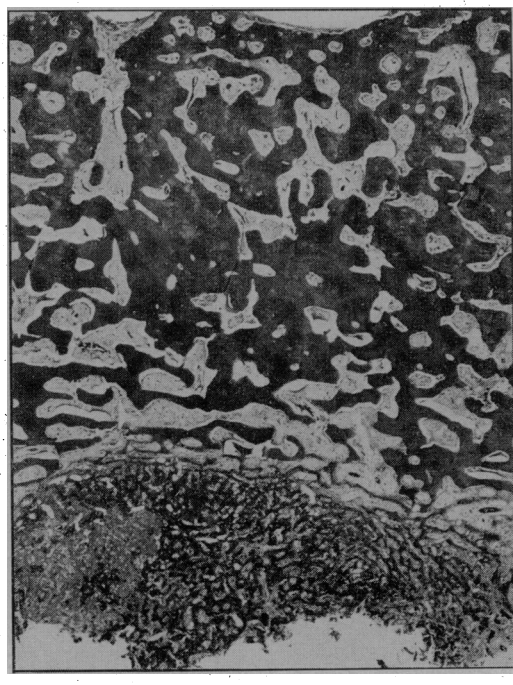


FIG. 6.

Case I.—A boy of 12 had suffered for one year from increasingly constant pain in the right hip and thigh. The lesion is slightly above the middle of the femoral shaft, and stands out clearly in thickened cortical bone (Fig. 1). When removed intact in its setting (Fig. 2), the nidus appeared as a dark red, gritty, friable tissue mass. The histologic pattern of the lesional area is shown in Fig. 5.

Case II.—A boy of 18 had complained, again for one year, of rather constant localized pain and some swelling in the lower part of the right leg. The lesion is in the lower third of the tibial shaft, and stands out fairly clearly as a roundish relatively radiolucent area in a setting of thickened cortex and spongiosa (Fig. 3). When removed intact in its setting (Fig. 4), the nidus appeared as a brownish globule of a friable but gritty tissue. The histologic pattern of the lesional area is shown in Fig. 6.

Case III.—A boy of 19 had suffered for two years from increasingly persistent and progressive pain in the right elbow region. In the X-ray film, the nidus stood out fairly clearly as a round focus of relative radiolucency set in an area of cortical thickening. When the nidus was removed intact in its setting, it was found to be a cherry red, granular, gritty tissue mass, 1 cm. in diameter.

This case illustrates that the nidus may fail to stand out against the surrounding thickened bone if insufficient kilovoltage is used in the exposure of the film. Indeed, radiographs taken under good medical auspices shortly before the patient came to us failed to reveal the nidus, and the observed thickening in the lower part of the humeral shaft was not ascribed to an osteoid-osteoma. However, when the lesion was coned down upon, and increased kilovoltage used in the radiography, the nidus came to stand out clearly, and all the data were harmonized for the correct diagnosis.

It should now be pointed out, however, that one also encounters cases in which, even under favourable radiographic conditions, a cortical nidus does not stand out as a clear-cut focus of increased radiolucency. When in the spongiosa at the articular end of a long bone, the nidus may present a relatively radiolucent or a relatively radiopaque shadow.

Case IV.—A woman of 49 had been suffering for two years from increasingly persistent pain in the left knee. An osteoid-osteoma was located in the spongiosa of the lateral condyle of the femur, at its lateral margin, just beneath the articular cartilage. The nidus appeared as a roundish, vaguely radiolucent area without evidence of perifocal reaction. Indeed, the presence of the nidus could be overlooked, as it was for a long time despite repeated X-ray examinations.

Case V.—In this case we are dealing with an osteoid-osteoma in the spongiosa at the upper end of a fibula. The patient was a boy of 19 who complained of pain of nine months' duration, localized to the outer aspect of the left leg, near the knee-joint. The nidus in this case contrasted with the one in the previous case, in that it stood out very well as a small, roundish radiopaque body, beyond which was a marginal crescent of densified spongiosa.

Case VI.—A boy of 16 had complained of pain in the right ankle for two years. The nidus, at the lower articular end of the tibia, appeared as a radiopaque body. The interesting question raised by this case was the differential diagnosis between osteoid-osteoma and osteochondritis dissecans. I have seen several instances in which an osteoid-osteoma presenting a radiopaque nidus at an articular end of a long bone was thought to represent osteochondritis dissecans until anatomic study of the removed tissue rectified that impression.

One or another tarsal bone (but especially the talus or the calcaneum) is not infrequently the site of an osteoid-osteoma. In relation to the calcaneum, the complaints are usually prolonged before the condition is recognized.

Case VII.—A medical student, 24 years of age, had suffered for sixteen months from progressive disabling pain in the right foot. Repeated X-ray examinations had been made but the findings had always been reported as negative, and the patient had consequently come to be regarded as a psychoneurotic. The X-ray picture taken a few days before surgical intervention showed the nidus located in the spongiosa toward the lateral surface of the calcaneum. It was small and round and not particularly conspicuous. It was surrounded by a rather narrow, irregular zone of radiopacity.

Case VIII.—A schoolteacher of 31 complained of pain in the right ankle, of three years' duration. During this time, she had had numerous radiographs taken of the ankle. The film that led to the surgical intervention shows an area of sclerosis in the calcaneum, beneath the subtalar joint, but the osteoid-osteoma nidus does not stand out particularly clearly within this general area of increased radiopacity (Fig. 10).

Only occasionally does one encounter an osteoid-osteoma in a carpal bone.

Case IX.—A man of 35 who complained of persistent, progressing pain in the right wrist, of one and a half years' standing. In addition, there was a point of exquisite tenderness over the semilunare, which showed a small, nidus-like focus of radiopacity. This case was particularly interesting because the radiopaque body seen in the X-ray picture represented an enostosis or "bone fleck" in which an osteoid-osteoma was developing.

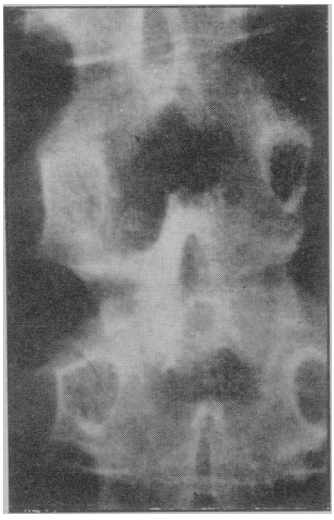
Occasionally, an enostosis (which by itself is painless) is misinterpreted as an osteoid-osteoma because the patient is complaining of pain in the bone or joint area in which the enostosis is present. The example just given of an osteoid-osteoma developing *within* an enostosis is the only one we have encountered. It seems to represent one of those oddities which complicate but add to the interest of problems of clinical diagnosis.

A metacarpal or a metatarsal bone, or a phalanx of a hand or foot is likewise occasionally the site

of an osteoid-osteoma. In a phalanx, the nidus may show up as an area of radiolucency, or as a roundish core of radiopacity as in Fig. 8.

As to the involvement of bones of the trunk, an osteoid-osteoma in a vertebra is not uncommon. We have also seen one case with an involvement of an innominate bone (Fig. 11), and one of a rib where the lesion was in the posterior end of the right 11th rib (Fig. 9). The patient was a boy of 15 who complained of pain in the right flank and costovertebral region for about one year. Sneezing, coughing and hiccoughing aggravated the pain, and, in time, the patient developed a dorso-lumbar scoliosis.

The body of a vertebra is nearly always spared, the lesion usually developing in some part of the vertebral arch, or in a process. On account of the difficulty in obtaining good radiographs of these processes, the clinical diagnosis is often difficult.



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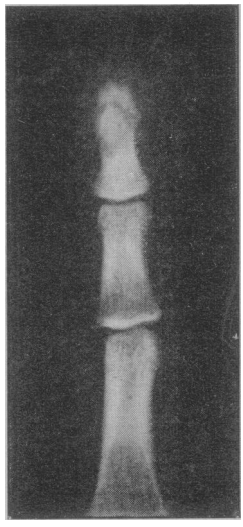

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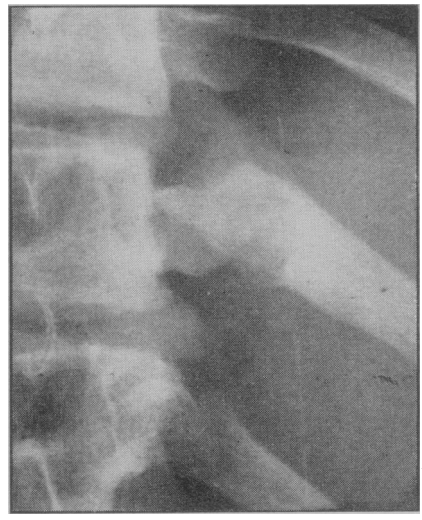

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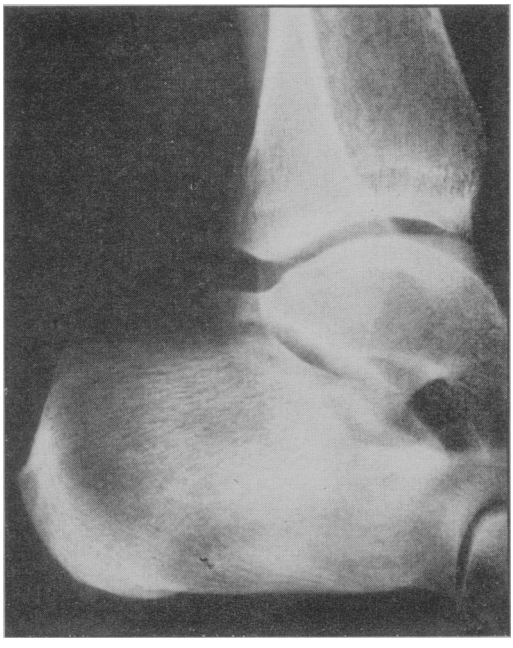

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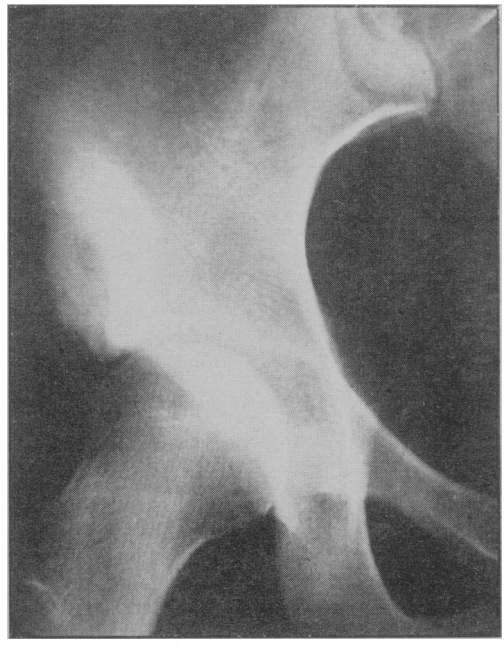

FIG. 11.

Case X.—The patient was a boy of 15 with pain in the upper part of the back, and an area of spot tenderness over the 4th dorsal vertebra, on the left side. He eventually developed kyphoscoliosis. After fourteen months, the back was explored. An osteoid-osteoma was found which involved the pedicle and the adjacent part of the lamina of the 4th dorsal vertebra on the left side.

Case XI.—The patient was a girl of 10 who complained of progressive disabling pain in the back for almost a year. The lesion, in the upper aspect of the right lamina of the 2nd lumbar vertebra, shows in the radiograph as a roundish area of relative radiolucency, almost completely surrounded by a zone of relative radiopacity (Fig. 7). At surgical intervention, the nidus was found to be globular, red, partly soft and partly calcareous, and the size of a cherry.

Case XII.—In this patient, a young man of 21, there was a five-year history of back pain of increasing severity. The lesion was at the base of the left transverse process of the 2nd lumbar vertebra.

Some clinical considerations.—From these case presentations, one can gather that an osteoid-osteoma may develop in almost any bone, though we ourselves have not observed it in a skull bone. About 25% of our cases involved a femur, and about 25% a tibia.

The lesion seems to be at least twice as common in males as in females. Fully 75% of the patients are between 10 and 25 years of age. Few indeed are below 5 or over 50 years of age.

The duration of complaints at the time of admission for removal of the lesion is usually between six months and two years. Histories of less than six months are the exception, and only an occasional patient in our series gave a history as long as five years.

Probably the main reason for the delay in the clinical identification of the condition is that, during the early stages, it may be difficult to demonstrate the osteoid-osteoma nidus radiographically even under favourable technical conditions. Another reason undoubtedly is that the general medical practitioner—the one most likely to see the patient during the earlier part of the clinical course—is not likely to think of the possibility of an osteoid-osteoma.

The principal complaint is of pain, and it is this that consistently leads the patient to seek medical attention. Occasional and mild at first, the pain usually increases in persistence and severity, and often becomes bad enough to interfere with sleep. Fortunately, most patients learn quickly that an adequate dose of aspirin almost consistently relieves the pain for several hours at a stretch. The principal clinical finding is local tenderness—often exquisite "point" tenderness. I shall not attempt to discuss such clinical questions as the rather common finding of referred pain and the occasional finding of inexplicable neurologic manifestations. However, I would like to point out that, ordinarily, immobilization of the part does not relieve the pain.

Pathogenesis.—The evolutionary sequence in the development of the osteoid-osteoma lesion has always intrigued me (Jaffe, 1945), but I must forewarn you that I still do not feel that I understand it completely. The genesis of the lesion is, of course, the same whether in a spongy bone area or in relation to cortex. However, it is much easier to line up what seems to be a series of developmental stages for cortical lesions.

At a site where an osteoid-osteoma has started to evolve, the osseous tissue comes to stand out from the surrounding bone. The contrast is created by the effects of increased vascularization and active reconstruction at the site. In consequence of this, the original osseous tissue in the area comes to be resorbed and simultaneously replaced by new bone laid down in a process of creeping replacement. The new bone presents a rather atypical histologic pattern, and its deposition is most striking around blood vessels. Sooner or later, the overlying periosteum becomes stimulated to deposit new bone on the outer surface of the cortex.

Eventually, the area under reconstruction becomes completely converted into a nidus of closely compacted trabeculæ of highly atypical osseous tissue. Between these trabeculæ, there are numerous thin-walled blood channels set in a sparse stroma of osteogenic connective tissue. From comparative study of many lesions, one gathers that, if an osteoid-osteoma nidus presenting such a histologic pattern is not removed and thus has a chance to evolve further, the nidal area continues to undergo reconstruction. Its trabeculæ of highly atypical bone are reworked and resorbed, and a much more productive nidus comes into being. In the foreground of such a nidus, we now have a substratum of very vascular osteogenic connective tissue in which osteoid and osseous tissue in varying proportions are being laid down.

In fact, if one took an osteoid-osteoma nidus in this advanced stage of evolution out of its setting, it might quite reasonably be misinterpreted, in some cases, as a tumour field from an osteogenic sarcoma. The neoplastic aura is given it by the evidence of active osteogenesis within it. In fact, it is this aura of neoplasia that I had in mind when I gave the lesion the name of "osteoid-osteoma" in spite of its consistent smallness and apparently self-limited nature.

Treatment.—Surgical removal of the lesion is all that is needed to bring prompt and lasting relief. The nidus and some of the neighbouring bone may be removed with a curette, but this removal should be thorough, since otherwise there is danger of recurrence of the complaints. The only disadvantage

of curettement is a purely scientific one—namely that the tissue may be so crumbled that, unless the pathologist or surgeon is thoroughly familiar with the condition, he may fail to pick out the significant fragments (representing the nidus) for histologic examination, and thus fail to obtain confirmation of the clinical diagnosis. In acquiring experience with the lesion, I have sought, whenever possible, to obtain the nidus intact in its setting. This remains the procedure best calculated to facilitate wider understanding of the condition while abolishing the complaint of the patient.

What would be the ultimate state of our patients had there been no surgical intervention at all, we do not know. It is noteworthy, however, that, among them, a few had had complaints for as long as four or five years before the lesion was removed and relief ensued. Nevertheless, it is conceivable that an osteoid-osteoma might, after many years, undergo spontaneous clinical arrest or even anatomic involution: it will be difficult to collect evidence on this point, because of the severity of the clinical complaints and the usual ready accessibility of the lesion for removal. On the other hand, we do know that if, in the course of the operation, the osteoid-osteoma is not removed or destroyed, the clinical complaints are either not relieved at all, or, if relieved, recur after a variable time.

As to the effect of irradiation therapy on an osteoid-osteoma, little is known. One interesting case in point was that of a boy of 15 when a humeral lesion which had troubled him for a year was diagnosed as an osteoid-osteoma. At that time, he had received a course of ten X-ray treatments (of 200 r each) against the lesion. After temporary aggravation, the pain was relieved for almost three years. His complaints then recurred, and, after they had persisted for about a year, the lesion was excised. During the entire preceding five years, the osteoid-osteoma had remained visible radiographically. On histologic examination of the tissue removed at operation, an osteoid-osteoma nidus was still clearly identifiable.

Finally, I shall describe a case (the only one I have seen) of a recurrence at the site of the original lesion. In 1938 at the age of 3, a girl was treated for what was supposed to be a "sclerosing osteitis of Garré" affecting the left tibia. In the tissue then removed, an osteoid-osteoma was observed. Unfortunately, the original radiograph is not available. In 1952, at the age of 17, the patient was readmitted on account of complaints of one year's standing, again referable to the upper part of the left tibia. The tissue now removed showed again an osteoid-osteoma nidus. It is difficult to decide whether this second lesion represents a recrudescence of the first or the formation of a new one at the same site. The case merely calls attention to the possibility of what one may neutrally call a "recurrence" of an osteoid-osteoma, after a thirteen-year interval of freedom from complaints between the original and the second lesion.

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