Case I

A girl aged 11 was admitted to hospital on December 13, 1937, with a history of three days' illness—typical acute osteomyelitis of the lower end of the right femur with abscess formation. The child was very ill, and had a temperature of 102° and pulse of 126. An incision was made down to the bone, which was not drilled (Mr. S. G. Davidson). On the 17th there was pain and swelling of the left tibia, followed on the 18th by pain and swelling of the right humerus. Uleron, 7½ grains, was given thrice daily for about a week, beginning on December 17. Three days after uleron was started the temperature began to fall and the general condition rapidly improved. There was no operative treatment for the tibia or humerus. The patient was discharged in April, 1938, with the wound in the right leg healed.

Skiagrams showed the typical appearance of osteomyelitis in the femur, tibia, and humerus. *Staphylococcus aureus* was found in the pus. The report from a blood culture was negative, but there must have been considerable blood infection, as proved by the appearance of two very definite secondary foci.

Case II

A boy aged 4 was admitted to hospital on July 15, 1938, with a history of three days' illness, and also of swelling and pain at the lower end of the femur. The appearance was typical of osteomyelitis; temperature 102°, pulse 160. The child was gravely ill, and it was considered that any operative procedure was not justified. Uleron was given every four hours and continued for three weeks, when the child's face began to look rather bluish and use of the drug was discontinued. Two days after uleron was started his condition began to improve, and his temperature fell to normal in a week.

Skiagrams showed periosteal thickening at the lower end of the femur and, later on, a small focus in the shaft higher up. Blood culture showed the presence of *Staph. aureus*. The child was discharged one month after admission apparently cured.

Case III

A male child aged 2 was admitted to hospital on June 23, 1938, having a history of two days' illness, with pain and swelling at the upper end of the left tibia and an abscess. He was very ill, his temperature being 104° and his pulse 160. Uleron, 7½ grains, was given every four hours. On June 27 the abscess was incised down to the bone, and on July 20 the whole shaft was easily removed subperiosteally. A small part of the circumference of the shaft was sawn off in all its length, boiled, and replaced in the periosteal bed. Pus and blood culture made on June 27 showed *Staph. aureus*, but on July 6 blood culture was negative. A small secondary abscess developed in the lower end of the right radius, but this cleared up quickly after being drilled. On August 19 the leg wound was very clean. The granulations were exceptionally fresh-looking, and the tibia was re-forming in very good position.

Case IV

A youth aged 16 was admitted to a nursing home on July 21, 1938, with pain and swelling of the lower end of the right femur. The temperature was 106° and pulse 120. The abscess was incised on the same day. Four drill-holes were made and a small piece of the cortex of the femur was removed. Uleron, 7½ grains, was given every four hours. Blood culture was positive to *Staph. aureus*. A second blood culture on August 1 was negative. This patient showed a very rapid improvement, but uleron had to be discontinued after eight days owing to skin irritation.

Skiagrams showed that a considerable length of the shaft of the femur was infected. On August 18 there was very little discharge from the wound and the patient's general condition was excellent.

Case V

A boy aged 6 was admitted to hospital on August 10, 1938, having a history of two days' illness. His temperature was 102° and the pulse 138. There was pain and swelling over the lower end of the right femur, and the abscess was incised. Two drill-holes were made in the lower end of the femur; acriflavine and vaseline gauze dressing were applied and the leg fixed in plaster-of-Paris. Uleron, 7½ grains, was given every four hours. The temperature fell to normal in eight days. The child was very comfortable, and up to the time of writing the dressing has not been changed. Examination of the pus showed the presence of *Staph. aureus*. Blood culture was negative. This case was not quite so severe as the others, and has made exceptionally good progress.

Commentary

These case notes have been made as brief as is possible, and any discussion of local dressings, splintage, and sun exposure has been omitted.

As I have pointed out, the series is a short one, and it may be argued that similar recoveries might have been obtained by treatment other than that adopted, but I feel strongly that all these cases have done exceptionally well, particularly Cases I and II, which seemed absolutely hopeless. All of them were definitely of the really severe septic osteomyelitis, and not of the milder type sometimes seen.

PELVIC HYDATID CYSTS AND OBSTRUCTED LABOUR

BY

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Perusal of the literature leaves little doubt that, in this country at least, obstructed labour is only rarely caused by a hydatid cyst. Such a case recently seen in the practice of the maternity department of the Cardiff Royal Infirmary prompted me to consult the hospital records. They revealed a similar case fifteen years ago, and so this opportunity is taken of describing both cases.

Case I

A 5-gravida aged 33 was admitted on October 24, 1923, having been referred by Dr. Burns of Abertillery. She had previously had four living children—three *per vias naturales* and one by Caesarean section—and no miscarriages. The patient had been unwell during the early months of pregnancy and a hydatid cyst had been removed from the anterior aspect of the uterus at the fifth month.

Examination revealed a full-term pregnancy, labour being obstructed by a large mass in the pouch of Douglas. Classical Caesarean section was performed and a full-term living child weighing 6 lb. was delivered. The uterine incision was brought together with mattress sutures and the abdomen explored. The uterus, especially its right lower aspect, was studded with small tubercle-like bodies. There was a large retro-uterine fluctuant mass in the pelvis, with a number of hard white masses resembling caseous glands attached to the omentum. On manipulation the pelvic mass ruptured, and what appeared to be pus and masses of endocyst were extruded. A large number of cysts of all sizes were attached to the omentum and peritoneal surfaces, and were removed. Further exploration revealed a hard calcareous mass attached to the under surface of the liver, but whether primarily there or in adherent omentum could not be ascertained. Removal of the mass was

considered inadvisable. Subtotal hysterectomy was then performed, with removal of the left appendages and what remained of the right, and the abdomen was closed.

Progress was fairly satisfactory until the twenty-fourth day after operation, when the patient suddenly collapsed and died, apparently from a pulmonary embolism. Permission for a post-mortem examination was not obtained.

Case II

An 8-gravida aged 35 was referred to the Infirmary by Dr. Roberts of Barry on February 5, 1938, as a case of labour obstructed by a pelvic tumour. She had had three miscarriages followed by four normal confinements, the last being two and a half years previously.

At the time of admission the membranes had been ruptured thirty-five hours and labour had been in progress sixteen hours. Abdominal examination revealed a full-term pregnancy, the foetus presenting by the breech in the left sacro-anterior position and the foetal heart rate being 138 per minute. Contractions were recurring every five minutes. On vaginal examination the cervix was found to be half dilated, with the breech presenting high up. There was a firm irregular mass in the pouch of Douglas about the size of a coconut which did not appear to be attached to the uterus and which was obviously obstructing labour. A tentative diagnosis of impacted ovarian cyst was made.

Classical Caesarean section was performed and the patient delivered of a full-term living child weighing 6½ lb. The uterine wound was closed and the abdomen explored. There were multiple cysts in the abdomen—eight to ten in number. The ovaries were not involved. One cyst attached to the upper surface of the bladder, and a second attached to the descending colon by adhesions, were removed for microscopical examination. Other cysts were intimately attached to the colon and mesocolon, and varied from the size of a walnut to that of a coconut. One such cyst was impacted in the pouch of Douglas, obstructing labour. As removal of all the cysts would have meant considerable dissection and have taken much time, it was thought advisable to close the abdomen. Convalescence was uneventful, and the patient was discharged twenty-five days after operation.

The diagnosis of hydatid cyst was confirmed microscopically. A blood count revealed an eosinophilia of 5 per cent. and the Casoni reaction was positive. The patient had always enjoyed good health, and inquiry failed to elicit any previous, family, or social history suggesting hydatid disease.

Pathological and Clinical Considerations

The two cases described were almost certainly examples of secondary echinococcosis. They serve to emphasize certain pathological and clinical aspects of hydatid disease which are as yet not sufficiently appreciated and which may be briefly outlined.

Following ingestion of the ovum the hexacanth embryo hatches in the upper part of the alimentary canal and, travelling via the radicles of the portal vein, lodges as a rule in the liver. Here, as a result of growth changes which need not be detailed, the typical univesicular hydatid cyst is formed, consisting of an outer ectocyst, or laminated layer, and an inner endocyst, or germinal layer containing specific fluid in which float the reproductive elements—the whole surrounded by a fibrous adventitia derived from the host. The scolices grow inside specialized brood capsules, and with their formation the cyst may be said to be fully developed. Such a cyst is typically found in a child or young adult; for in the majority of cases infestation takes place in childhood, so that the cyst is nearly as old as the host.

In nearly all instances it is the onset of complications that determines the first clinical manifestation of hydatid disease. In the great majority of cases these complications

depend on the previous escape of fluid from the cyst—varying from a slight leak to a frank rupture—and commonly occur after the third decade of life.

Rupture of the cyst may take place into the subcutaneous or muscular tissues, into the bile ducts, bronchi, alimentary canal, or urinary tract; into a serous cavity such as the peritoneum, pleura, or pericardium; or into the chambers of the heart or large veins. Each presents its own characteristics, but according to Dew (1930-1) the sequelae may be grouped as follows:

General—applicable to all types:

1. Immediate: Anaphylaxis.
2. Delayed: Secondary echinococcosis.

Special—applicable particularly to rupture into a natural channel:

1. Immediate: Mechanical effects.
2. Delayed: Suppuration in the cyst.

Of these sequelae it is only necessary to consider briefly anaphylaxis and secondary echinococcosis.

ANAPHYLAXIS

The appearance of anaphylactic phenomena following rupture or puncture of a hydatid cyst has been noted many times. The most common of these manifestations are urticaria, dyspnoea, cyanosis, abdominal pain, vomiting, syncope, and delirium. The severity of the anaphylactic reaction varies within wide limits, but it is probable that were more accurate histories obtained clinical evidence of anaphylaxis would often be revealed.

SECONDARY ECHINOCOCCOSIS

For many years the majority of clinicians regarded multiple cysts as evidence of multiple primary infestations, but it is now known that they are nearly always manifestations of secondary echinococcosis following rupture of the primary cyst and the implantation of daughter cysts or scolices. There are many clinical examples of this phenomenon, but only two—the localized and intraperitoneal types—need be mentioned.

Slight leakage of fluid from a single cyst, following puncture or mild trauma, may give rise to the formation of multiple secondary cysts within the confines of the adventitia, and is one of the commonest types of reactive daughter-cyst formation. Dew is of the opinion that probably all cases of endogenous daughter-cyst formation are best regarded as a special type of localized secondary echinococcosis.

Multiple secondary cysts of the peritoneal cavity are relatively common, and are always due to leakage of reproductive elements from a primary cyst of the liver or, rarely, the spleen or kidney. These secondary cysts become completely surrounded by the peritoneal epithelium, so as often to give the impression that they have developed in an extraperitoneal situation. They are of slow growth, with a latent period of from five to ten years before they produce symptoms. They are prone to further rupture, with the production of more cysts, so that ultimately the peritoneal cavity may be almost filled with hydatid cysts in all stages of development. The original primary cyst may be completely evacuated, so that after a lapse of years it may be exceedingly difficult to detect the primary site. More commonly, however, the rupture becomes sealed off by adhesions, and the residual germinal elements produce multiple daughter cysts—one type of localized secondary echinococcosis.

The multiple intraperitoneal cysts present in each of the cases described were undoubtedly produced by rupture of a primary hydatid cyst as outlined above. In Case I

the calcareous mass attached to the under surface of the liver would appear to have been the remains of the primary cyst. In Case II the primary cyst was not located. In neither case was a history suggestive of anaphylaxis obtained, so that the time of rupture of the primary cyst cannot be known; but it is likely that in each case multiple cysts had been present and unsuspected in the peritoneal cavity for several years.

In Case I the first manifestation of hydatid disease appears to have been in the earlier months of the pregnancy, when the patient began to feel unwell. Presumably in consequence of an examination laparotomy was performed and a hydatid cyst removed from the anterior part of the uterus. In Case II hydatid disease was not suspected until the occurrence of an obstructed labour due to a cyst being impacted in the pouch of Douglas. It is noteworthy that in both cases, although hydatid disease had probably been present for many years, it was not discovered until the fourth decade of life, and then only on account of an associated pregnancy.

Literature

References in the literature to the ubiquitous hydatid cyst in relation to pregnancy and parturition are few. Franta (1902) collected all the recorded cases and dealt with them in three papers. The first was concerned with dystocia due to hydatid cysts in the pelvis, and recorded thirty-six cases. The second contained an account of twenty-two cases in which operation for removal of the cyst was performed in pregnancy. The third dealt with the influence of pregnancy, labour, and the puerperium on hydatid cysts. I am aware of only four recorded instances in this country. Gemmell (1899) described a case of hydatid cyst in the omentum obstructing labour which was treated by abdominal section and removal in the puerperium. Blacker (1908) published a case of hydatid cysts complicating pregnancy in which Porro-Caesarean section was performed. Andrews (1908) also described a case in which pregnancy was complicated by the presence of a hydatid cyst in the pelvis. The remaining case is that of Bell (1925), whose patient had an obstructed labour necessitating craniotomy, the cyst subsequently being removed during a second pregnancy.

Summary

1. Two cases of obstructed labour due to a hydatid cyst are described because of their comparative rarity.
2. Both cases were examples of secondary echinococcosis with multiple cysts in the peritoneal cavity.
3. In neither case was there a history of anaphylactic manifestations such as frequently accompany rupture of the primary cyst.
4. Although hydatid disease had probably been present for several years, it had not been suspected. In the first case it was discovered at the fifth month of pregnancy, and in the second during labour.

I wish to express my thanks to Sir Ewen Maclean and Dr. M. D. Arwyn Evans for permission to publish these cases.

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EPIDEMIC MYALGIA FIVE CASES IN ONE HOUSEHOLD

BY

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The following account of an outbreak of epidemic myalgia may be of some value, as it provides a possible differential diagnosis in cases of appendicitis, pleurisy, cholecystitis, and muscular rheumatism.

Survey of Literature

Daae (1874) in Norway is first credited with describing the disease, which, he says, is distinguished by pain in the chest and back, febrile disturbance, and relapses. Gradually it became recognized as a clinical entity, and by 1888 the name "devil's grip" had been associated with it in America. From this time it attracted little attention until 1930, when Sylvest (1930, 1931) gave a full account of an epidemic which occurred on the island of Bornholm, in which 10 per cent. of the population were attacked. He described the onset as sudden, with pain in the hypogastrium and nausea, the pain shifting to the chest and shoulders after a few hours. These symptoms are associated with fever, from 99° to 105° F., and the attack lasts a few days. Relapses are frequent. Sylvest noticed a seasonal incidence in summer and early autumn. The sexes are equally affected, and, although no age is exempt, most cases occur between the ages of 5 and 15. There appeared to be an incubation period of two to four days, and examination of the blood showed an eosinophilia.

Since 1930 there have been a few reports in the literature. Rector (1935) records nineteen cases in which the patient was under 8 years old, and he suggests the use of quinine sulphate in cases that do not clear up quickly. Smith (1937) gives an account of an epidemic at Rugby School, in which there were myalgic pains, chiefly in the back of the neck.

In 1934 the disease received more attention owing to several outbreaks in American towns. MacDonald, Howell, and Cooper (1937) investigated an outbreak in Cincinnati; clinically they found the same picture as did Sylvest, but there were no patients over 15 years of age. They point out that constipation was frequent and relapses after a few days were common features. These workers found no eosinophilia, but some of the patients had a leucopenia. Bacteriological investigations of the cerebrospinal fluid were negative, as also were blood cultures. The Wassermann reaction was negative in all cases. Cooper and Keller (1937) tried to find a specific organism in washings of the throat and nose, but the only constant growth was a small colony type of *Streptococcus viridans*, to which they attached little importance. Later work with animals suggested a filterable virus as the cause, with a possible incubation period of eight to ten days. F. Wolter (1937), investigating a series of outbreaks in Germany, adds the fact that the urine is usually very dark in colour, and this, he suggests, is due to the presence of creatinine, which he attributes to the changes in the muscles. He discusses the connexion between epidemic myalgia and epidemic poliomyelitis, and points out that there is a similar seasonal and climatic incidence.