A Case of Multiple Pulmonary Infarctions Occurring in an Ambulant Male, and Associated with Rectal Lesions*

DANIEL J. STONE, M.D.† and FRANCIS J. LOVELOCK, M.D.††
Bronx, New York

The problem of pulmonary embolism with infarction continues to fascinate the pathologist and clinician alike. With the advent of the pioneer work of Homans,† Allen‡ and others.¶ The emphasis, and rightly so, has been on the dangers of deep vein thromboses, particularly in the calf, femoral and iliac areas. Its relationship in the debilitated, the surgical and cardiac patient, to pulmonary artery embolization is too well documented to require discussion.

Less well publicized, are the occasional cases of pulmonary infarction, occurring in ambulant and apparently well individuals. Except for one or two scattered case reports, this has received little attention. The rectum and its venous drainage as a possible source of pulmonary emboli has been mentioned as such in only one relatively recent study.§

It is the purpose of this paper to discuss the problem of the rectum as a source of embolization and to report a case which suggests that possibility.

A 33-year old Negro male was admitted on January 2, 1948 for pleuritic pain (in the right side of the chest) of one month's duration. He had no symptom of pulmonary disease prior to October 1945, when he developed an acute attack of fever and wheezing while on Saipan. A diagnosis of bronchial asthma was made, and he was discharged in January 1946. He continued to have mild bouts of asthma, occurring every two or three months, which were treated symptomatically, and were never associated with cough or sputum.

The present illness began suddenly on December 2, 1947, with malaise, fever and severe chest pain localized in the area of the right nipple, axillary and scapular regions. The pain was knife-like in character and aggravated by the slightest respiratory movement. Shortly after onset,

*From the Medical Service of the Veterans Administration Hospital, Bronx, New York. Reviewed in the Veterans Administration and published with the approval of the Chief Medical Director. The statements and conclusions published by the authors are the result of their own study and do not necessarily reflect the opinion or policy of the Veterans Administration.
†Assistant Chief, Chest Medical Section.
††Chief, Chest Medical Section.
four to five teaspoonfulls of bright red blood was expectorated. On December 5 he was admitted to another hospital. A diagnosis of pneumonia was made, and he was treated with penicillin, becoming symptom-free and afebrile over a 10-day period. On the 20th of December, five days after ambulation had been started, the same symptom-complex recurred with pleuritic pain in the right lower chest and with x-ray evidence of a pleural reaction at the right base. He was retreated with penicillin for a seven-day period and was discharged from the hospital, symptom-free on December 30, 1947. He was admitted here on the second of January 1948 merely for a check-up examination.

Past history included bleeding internal hemorrhoids operated on in 1942 and 1944. One year prior to admission he had a spontaneous discharge of pus from the rectum. Five months before admission he noted occasional serosanguinous discharge from the rectum and pain unrelated to defecation. He was treated for a chancere in 1939, and retreated in 1944 because of positive serology.

On admission to this hospital his temperature was 99.9 degrees F., pulse 90, and respiration 20. He was a thin, asthenic Negro not appearing acutely ill, but somewhat malnourished and fatigued. There was right lower dorsal scoliosis. Over the right lower lobe there was dullness, diminished fremitus and breath sounds and post-tussic fine rales. Blood pressure was normal and cardiac examination was negative. Peripheral arterial pulsations were all normal. No edema, discoloration or heat was noted in the lower extremities. Homans' sign and calf tenderness were absent. The liver edge was palpable just beneath the costal margin, and was firm and non-tender. Rectal examination revealed a ring of tender external hemorrhoidal tags. Above the sphincter, several observers noted a roughened and somewhat nodular area on the posterior surface of the rectum, which was slightly tender. No rectal discharge was noted on admission. The remainder of the physical examination was within normal limits.

Sedimentation rate was 23 mm./hr. (Cutler method). A Kahn titre of less than 10 units was reported. X-ray films and fluoroscopy revealed a rather dense pleuritic reaction at the right base posteriorly with a mottled infiltrate in the same area. An old ununited fracture of the axillary segment of the right, fifth rib was noted.

Four days after admission (on January 6, 1948) he developed severe left pleuritic chest pain requiring administration of codeine. Physical examination revealed suppression of breath and voice sounds in the left axilla. He was apprehensive, with pulse rate of 100 and normal temperature. With the onset of this episode, dark bloody sputum, mixed with small clots was noted and he continued to raise small amounts of dark red blood and rusty sputum for the next four or five days. Chest x-ray film, including oblique projection revealed in addition to the previous findings, a small pleural effusion at the left base and a definite circumscribed parenchymal infiltrate in the base of the left lung (Figure 1).

A careful re-examination of the peripheral veins was negative. Because of the previous rectal findings, re-examination was done and the previously noted ragged area just above the sphincter was now distinctly tender. A cord-like projection was felt on the posterior wall of the rectum in the same area. Temperature became elevated and varied around 102 degrees F. for the next four days.

It was felt that the evidence was sufficient for a diagnosis of multiple
pulmonary emboll with infarction and the patient was treated with anticoagulants. Penicillin was added because of the rectal inflammation. The white blood count was 11,400 with moderate polymnucleosis. Liver chemistry was normal except for a transient elevation of the icterus index to 16 (on the first day of the acute episode) with later return to normal.

Numerous sputum examinations were made throughout this period, and they were negative for pyogens, fungi and acid fast bacilli. Numerous stool specimens were studied and cultured for parasites, including Schistosoma, and were entirely negative. Three blood cultures were likewise negative.

The patient maintained adequate prothrombin levels and was treated for one month on that regimen, until ambulation was effected. The pleuritic pain subsided in several days and sputum became thin and finally completely disappeared. Chest x-ray films revealed gradual clearing of the processes in both lung fields. Daily examination of the extremities was done for three weeks with no evidence of thrombosis at any time. He remained afebrile and essentially well throughout the rest of his hospital stay.

Following subsidence of the acute pulmonary process, attention was redirected toward the rectum and he was proctosoped on the 28th hospital day, when preliminary digital examination again revealed an induration along the posterior wall of the rectum, roughly 6 cm. above the anal opening. Because of marked tenderness in that area, a limited proctoscopy was performed and there was noted an area of irregular and shallow ulceration along the lower 6 cm. of rectal mucosa. The proctoscope with difficulty was inserted to 12 cm., where similar but smaller ulcerations were noted. A small area of muco-sanguinous material was observed. Biopsy was postponed because of the marked pain produced by instrumentation. It was felt that acute proctitis existed. Stool studies for parasites and cultures for amebae were made at that time and were negative, as was complement-fixation tests for amebiasis. The possibility of an acute proctitis secondary to lymphopathia venereum was raised and studies were made with this in mind. A complement-fixation test for lymphopathia venereum on March 6, 1948, over one month after admission, showed a doubtful reaction at a 1:5 dilution and a negative

FIGURE 1

FIGURE 2
reaction at 1:20 dilution. The Kahn test remained at less than 10 K.U. A Frei test done at this time was markedly positive. At no time did he have inguinal lymphadenopathy and he did not recall a recent penile lesion. Because of the possibility that he had lymphathia venereum proctitis, he was treated with sulfadiazine for one month with gradual subsidence of rectal pain and mucoid rectal discharge. Approximately 70 days after admission (and two weeks following discharge) he was resectoscoped to a distance of 10 inches. No ulceration was noted, but at the site of previous ulcers, dull, depressed atrophic patches, suggestive of scars, were noted. The mucous membrane was somewhat friable and bled on touch. The previously noted indurated granular areas above the anal opening were still present, but no longer tender. It was the impression that these latter areas were the residuals of a periphlebitis. Smear and cultures from these lesions were negative. Barium enema on March 5 revealed slight irregularity of the rectal mucosa. A repeat study in April 1948 was interpreted as normal.

When last seen in May 1948, the patient had no rectal or pulmonary symptom and his chest x-ray film revealed pleural thickening at both bases (Figure 2).

Discussion

Anatomy.7 The hypogastric or internal iliac vein drains much of the pelvic viscera, and it in turn joins the systemic circulation by uniting with the external iliac vein at the pelvic brim. Among tributaries which join this vein are the internal pudendal with its origins outside the pelvis and the middle hemorrhoidal veins originating in the pelvic viscera. The hemorrhoidal plexus surrounds the rectum and consists of an internal portion lying in the submucosa immediately above the anal opening and an external portion in the muscularis. This internal portion communicates freely with the external plexus and in turn with the vesical plexus. The inferior hemorrhoidal veins drain the lower portions of these plexuses by connection with the internal pudendal vein. The middle portion is drained by the middle hemorrhoidal vein and the latter joins the hypogastric vein. Only the upper portion of the rectum drains into the portal system, but because of the plexus arrangement, there is free communication between the portal and systemic circulations.

Four criteria must be satisfied to establish the hemorrhoidal plexus or rectal area as a source of systemic embolization; namely, anatomical, clinical, pathological and experimental.

1) From the discussion of the anatomy of this area (vide supra), it is clear that with the exception of the most superior portions of the rectum, there is free intercommunication between the rectal venous circulation and the systemic circulation. On morphologic grounds, there is no reason why the rectum could not serve as a source of venous systemic embolization.
2) If the rectum is to be clearly implicated as a source of pulmonary emboli, it would appear reasonable to demand some clinical and pathologic association of hemorrhoidal disease with pulmonary artery embolization. Since thrombotic hemorrhoidal disease is so common, pulmonary embolism should be a common clinical occurrence, provided there are no special factors tending to prevent it. With the exception of King’s report of several cases of pulmonary infarction possibly associated with rectal lesions, a review of the literature and common clinical experience reveal no definite association of pulmonary embolism and rectal pathology. This may possibly be explained, in part, by the anatomy of the hemorrhoidal veins. They are in the nature of varicosities and like those in the extremities, would probably not often be the source of embolism. An adequate explanation, however, of the absence of embolism in association with hemorrhoids is not at hand.

3) There is scarcely any pathological evidence to implicate the rectum as a source of pulmonary embolism. Allen, Spain, Neuhofer and others, in reviewing the sites of systemic emboli, do not mention the rectum as a possible source. Moran has published post-mortem material in which he lists a total of three patients in whom he felt hemorrhoidal thrombi were the source of pulmonary emboli. Unfortunately no details are given in this paper and there is no indication as to how complete the necropsies were. The pathological proof is therefore open to question in that series.

The failure of the pathologic reports on pulmonary infarction to implicate the rectum as a source of embolization may be due to the fact that the rectum and its circulation is not examined carefully in routine autopsies. Even in those cases of pulmonary embolism where the source was not obvious, examination of the rectal circulation was not made.

To prove pathologically that the rectum was the source of any given pulmonary embolus, one would require absence of thrombi in the usual locations, such as the deep veins of the extremity, and in addition, the presence of thrombosis in the inferior or middle hemorrhoidal venous plexus, and histologic similarity of the embolus in the pulmonary artery to the original hemorrhoidal thrombus.

The enormity of the above task in routine post-mortem work makes it understandable that the rectal circulation has been ignored in studies of thromboembolic disease.

It is of interest that in a number of pathological studies, the source of the pulmonary emboli were not found in a high percentage of cases; for example, 42 per cent of 200 cases (84 cases), in the series of Spain and Moses. In many of these studies it is
not made clear how often failure to find the source was due to lack of a complete examination, including leg dissection. It seems reasonable to suppose that in cases of pulmonary emboli in which examination of the usual sites of thrombi is negative, a careful search of pelvic veins, including the hemorrhoidal veins, might uncover their source.

4) Experimental criteria: A careful search of the literature reveals no record of any attempt to demonstrate the possibility of pulmonary embolization from experimentally induced hemorrhoidal thrombi. This might prove a fruitful field of investigation.

This case is presented as an instance of recurrent pulmonary infarctions in an ambulatory patient. There was no clinical evidence of thrombosis in the peripheral veins or the heart. There was, however, a severe inflammatory process in the rectum, associated with hemorrhoidal disease, which conceivably could have been the source of the emboli. The nature of this inflammatory process remains obscure. In view of the positive Frei test and the good clinical response to sulfadiazine therapy, it is possible that this process represented a lymphopathia venereum proctitis. A review of the literature on this subject does not reveal any reported instance of the association of pulmonary infarction and lymphopathia venereum.

In the series of 10 cases of pulmonary embolism occurring in ambulant and well Army personnel, reported by King et al., it is of great interest that in two cases pulmonary infarcts were associated with thrombosing, acute hemorrhoidal disease. Phlebography of the lower extremities in both these cases revealed no evidence of thrombosis.

It is anticipated that objection to the concept proposed in this paper may be made on the ground that neither this patient nor others referred to were studied by phlebography. It must be admitted that phlebograms here might have demonstrated thrombosis in the veins of the lower extremities. It is recognized, however, that the diagnostic value of phlebography in diagnosis of obscure sources (clinically inapparent) of pulmonary emboli is questionable. Allen found negative phlebograms in one-third of his cases of obvious clinical thrombotic disease of the lower extremity. Conversely, it is known that erroneous diagnosis of thrombosis may be made on phlebograms either because of veno-spasm due to the contrast media and venipuncture, or because of artifacts resulting from position of the extremity.

SUMMARY

An unusual case of multiple pulmonary infarctions occurring in a presumably well, ambulant and active young male has been
presented. Clinically it was striking that the embolic disease occurred in association with an unusual inflammatory process involving the hemorrhoidal veins which may have been due to the virus of lymphopathia venereum. It is recognized that this association may be coincidental, but it is suggested that the hemorrhoidal veins may have been the source of these emboli since no other was demonstrated.

The hypothesis presented above merits consideration, and it is hoped that in some small measure this report will stimulate clinical, pathological and experimental investigation of the rectal circulation in relation to pulmonary embolism.

RESUMEN

Es presentado un caso raro de infartos pulmonares múltiples en un varón joven ambulante, activo y al parecer sano. Clínicamente llamó la atención el hecho de que la enfermedad embólica ocurrió asociada a un proceso raro de inflamación de las venas hemorroidales, que podía ser causado por el virus de la linfopatía venérea. Se reconoce que tal asociación puede ser una coincidencia, pero se sugiere que las venas hemorroidales pueden haber sido el origen de estos émbolos, ya que no se demostró ninguno otro.

La hipótesis que se presenta merece consideración, y es de esperarse que, aunque sea en forma limitada, este informe estimule la investigación clínica, patológica y experimental de la circulación rectal en relación a la embolia pulmonar.

RESUME

Les auteurs rapportent un cas d'infarctus pulmonaires multiples, qui sont apparus chez un jeune homme bien portant, valide et en parfaite activité.

Cliniquement, il était frappant de constater que ces embolies s'associaient avec un processus inflammatoire atténuant les veines hémorroidaires, dont la cause semblait être le virus de la maladie de l'adéno-lymphoïdite. Les auteurs reconnaissent qu'il peut s'agir là d'une associations de coincidences mais ils suggèrent de considérer les veines hémorroidaires comme la source de ces embolies en l'absence de toute autre origine reconnue.

Cette hypothèse demande à être prise en considération, et les auteurs souhaitent que leur rapport puisse encourager des recherches cliniques, anatomiques et expérimentales sur les relations entre l'état des vaisseaux du rectum et les embolies pulmonaires.

REFERENCES

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