The Value of Registries for Rare Diseases: Bacterial or Mycotic Aortic Aneurysm

Running title: Hinchliffe et al.; Bacterial aortic aneurysm

Robert J. Hinchliffe MD, FRCS1; Janet T. Powell PhD, MD, FRCPath2

1St George’s Vascular Institute, London, United Kingdom;
2Imperial College (Charing Cross Hospital), London, United Kingdom

Address for Correspondence:
Janet T. Powell PhD, MD, FRCPath
Imperial College (Charing Cross Hospital)
St Dunstan’s Road
London W6 8RP
United Kingdom
Tel: +44-208-8467312
Fax: +44-208-8467330
E-mail: j.powell@imperial.ac.uk

Journal Subject Code: Cardiovascular (CV) surgery:[35] CV surgery: aortic and vascular disease

Key words: Editorial, aorta, aneurysm, registry, mycotic aneurysm
Bacterial infections of the aorta are rare. The label mycotic aneurysm is misleading, since mycoses or fungal infections of the aorta are much rarer still. The management of bacterial infections of the aorta has always been considered difficult, largely because they frequently herald aortic rupture and the outcomes of traditional surgery have been poor. Historically these patients were managed by surgical resection of the aneurysm, debridement of the infected tissue and revascularisation using in situ or extra-anatomic techniques with either prosthetic or less commonly autologous grafts. However, the results of such surgery have been disappointing with many patients being considered unfit to withstand the rigours of such major intervention and therefore palliated. Others have died as a result of the attempted curative surgery or had late infectious complications.

The creation of an international registry by Wanhainen and colleagues is an important step forward in improving patient management. It builds on case series reported by pioneering centres in the endovascular management of aortic disease that demonstrated it was possible to use an endovascular strategy in the management of aneurysms attributed to bacterial infection. These reports demonstrated the technical feasibility of the techniques but left many unanswered questions.

The management of rare diseases do not lend themselves well to assessment using standard surgical technique development tools. Prospective registries in the endovascular management of other uncommon aortic diseases including thoracic aortic dissection and aneurysm have proven utility. However, there are major drawbacks with such registries including reporting bias, missing data and selective loss to follow-up. Retrospective registries have additional limitations.

The primary consideration in understanding the role of endovascular interventions in
bacterial aneurysms is to assess feasibility. Any comparison of outcomes with historical series using standard surgical approaches must be tempered by the progress in antibiotic therapy and intensive care management. The report by Wanhainen and colleagues in this issue of Circulation demonstrates the feasibility of the endovascular approach across different centres in Europe, but prospective data will be needed to progress the management of bacterial aneurysms. It is interesting to note that few of these endovascular procedures were used as a ‘bridging procedure’ to definitive surgery, quite possibly because infections have apparently been arrested or controlled with antimicrobial therapy. An alternative explanation is that some or all of these patients were considered too frail to undergo major surgery or that possibly surgeons believe that the results of open surgery are no longer superior to an endovascular approach.

We are assured by the authors that “only a handful” of patients were treated with open repair but they do not provide numbers or outcomes of those treated with open surgery or no surgery at all. What proportion of patients presenting with bacterial aneurysms to the contributing centres were treated with endovascular interventions and can we be reassured that those patients treated with antibiotics alone or ‘palliated’ have such terrible outcomes?

The authors acknowledge that there are no standard diagnostic or reporting criteria for the diagnosis of infections of the aorta. Instead the authors used a panel of three criteria (biomarkers) for aortic infection used in everyday clinical practice (judged locally and confirmed in the ‘core lab’). However, it is quite plausible that some of these patients had alternative diagnoses such as an inflammatory aneurysm or connective tissue disorders. Further work will be required to develop consensus guidelines on the diagnosis of bacterial aneurysm (as for infective endocarditis and osteomyelitis of the diabetic foot).

Because this was a retrospective registry it was not possible to standardise clinical and
imaging follow-up. In a prospective registry it should be possible to strengthen the quality of the
data by standardising diagnosis and follow-up with pre-specified criteria and regimens.
Specifically, there were no data on the duration of pre-operative antibiotics, how patients were
followed-up and how missing data (including loss to follow-up) were handled. Were data
available for clinical (including microbiological and laboratory testing) and imaging follow-up
outcomes? Complications such as endoleak not attributed to infection are not reported. In such
cohorts it is quite possible that there may have been selective loss to follow-up.

Although the overall prognosis associated with endovascular treatment of bacterial
aneurysm appeared quite poor, the data are hard to interpret given the heterogeneous nature of
bacterial aneurysms with respect to site within the aorta, the presence of various causative
bacteria (and one Candida infection) and the evolution of endovascular technologies over the
14-year study period. Specific groups of patients appeared to be particularly vulnerable to
complications and premature death. The presence of gas on a CT scan resulted in a particularly
poor long-term survival (5yr survival 36%).

The authors also suggest that patients with a Salmonella culture had better outcomes than
those without a positive culture or culture of other bacteria. This requires confirmation in a
separate study, since it is possible that this observation was due to chance alone: the regression
analysis with 123 patients used 19 variables, so a much stronger p value would be needed to
confirm a statistically reliable association.

In summary, these data emanating from the registry on endovascular procedures in
aneurysms of bacterial etiology are a useful addition to the literature. However the registry has a
number of important limitations that hamper clinical interpretation of the data. Further work is
required to elucidate important information in regard to the diagnosis of bacterial aortic
aneurysm, the timing of intervention and to understand which patients may be better managed using an endovascular strategy. It is likely that a well-constructed prospective registry with clearly defined diagnostic criteria and standardised follow-up will be able to answer some of the outstanding questions.

Conflict of Interest Disclosures: None.

References:


The Value of Registries for Rare Diseases: Bacterial or Mycotic Aortic Aneurysm
Robert J. Hinchliffe and Janet T. Powell

Circulation. published online November 5, 2014;
Circulation is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 2014 American Heart Association, Inc. All rights reserved.
Print ISSN: 0009-7322. Online ISSN: 1524-4539

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://circ.ahajournals.org/content/early/2014/11/05/CIRCULATIONAHA.114.013367

Permissions: Requests for permissions to reproduce figures, tables, or portions of articles originally published in Circulation can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

Reprints: Information about reprints can be found online at: http://www.lww.com/reprints

Subscriptions: Information about subscribing to Circulation is online at: http://circ.ahajournals.org/subscriptions/